Changes in gait which occur before and during the adolescent growth spurt in children treated by Selective Dorsal Rhizotomy

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ABSTRACT
This paper presents long term follow up results from 17 children (6 girls, 11 boys, GMFCS levels II to IV), treated by means of selective dorsal rhizotomy (SDR). The particular focus is on the effect of the adolescent growth spurt on patients who had previously undergone SDR. The children were all assessed using 3D gait analysis, in combination with clinical examination at three time points - before SDR surgery (PRE), after SDR surgery when pre-adolescent (POST1) and post-adolescence (POST2). The total follow up period to POST 2 was 8y6m for girls and 9y5m for boys. All children maintained or improved their GMFCS level. Positive changes in ranges of motion and gait were observed at POST1 and these were generally maintained over adolescence to POST2. The mean Gait Profile Score (GPS) had improved by 3.2 points (14.7 to 11.5) at POST1, with a non-significant deterioration of 0.3 over the adolescent growth spurt. These positive results reflect the total package of care for the children, involving careful pre-operative selection by a multidisciplinary team and post-operative management including intensive physiotherapy and maintenance in tuned ankle foot orthoses. 59% of children had some additional orthopaedic surgery, mostly bony procedures. The overall benefits arising from their management need to be considered in the light of the likely deterioration experienced by this patient group. The results of this study support the use of SDR as part of a management strategy for carefully selected children with cerebral palsy with the aim of optimizing gait at skeletal maturity.
KEYWORDS: Cerebral palsy; Selective Dorsal Rhizotomy, Gait, Adolescence

INTRODUCTION

Selective Dorsal Rhizotomy (SDR) was refined and popularized by the team of Dr Peacock, in South Africa (1), to reduce spasticity in children with cerebral palsy. The procedure has been in use for over 30 years, with a substantial body of literature from centres publishing their local outcomes. The earliest patients treated are now adults, making it possible to assess the long term impact of SDR performed in childhood.

Despite substantial clinical experience of SDR there are very few randomized controlled trials, the main three (2,3,4) being summarized in the meta-analysis published by McLaughlin et al in 2002 (5). None of these studies used instrumented 3D gait analysis, and so were not able to report gait changes in detail. The follow up times are also relatively short at between 9 and 24 months.

The Oswestry SDR programme began in 1995. All children treated in Oswestry are assessed pre-operatively by means of a full lower limb clinical examination and gait assessment. These measures are then repeated 12 months post-operatively, and the short term outcome data have been presented elsewhere (6). A number of other centres have also reported short term outcome data, including pre and post-operative gait analysis (eg 7,8,9). Common features are seen in data from different centres. Following SDR children typically have greater passive extension at hip and knee and more dorsiflexion at the ankle. These changes are translated into better extension and foot posture during gait, along with a higher rate of knee flexion in swing leading to greater peak knee flexion. Global measures of gait function and quality (GMFCS, GGI) also show improvements. The only consistent negative change reported is increased anterior pelvic tilt.

Long term follow up data are more sparse, and rarely include gait analysis as an outcome measure. When SDR began to be offered 3D gait facilities were not commonly available. The team in Cape Town (10,11) have published their long term outcomes (up to 20 years post op) using a simple 2D gait analysis technique. They showed that key post-operative improvements, such as improved normalized step length and knee range of motion, continue to be apparent 20 years after SDR.

Other long term follow up studies use outcome measures such as GMFM, GMFCS and Ashworth scales (eg 12, 13, 14). Improvements seen after treatment are generally preserved, though studies lack the specific detail of gait patterns available from gait analysis. Beneficial changes seem to occur in the first 5 years after treatment, followed by a plateau period (13). Tedroff et al 2011 (15) reports maximum improvements at 3 years. However, whilst spasticity reduction was largely preserved, by 10 years joint range of motion and GMFM had deteriorated. Despite this GMFM values were still significantly better than pre-operative measurements.

In Oswestry, after the initial follow up visits at 1 and 2 years, we review our patients at fixed developmental milestones. In particular we are interested in observing children over the adolescent growth spurt, a period of rapid physical change commonly considered to pose a challenge to function (particularly gait) in cerebral palsy. Our long term follow up assessments are therefore performed at 10 and 16 years of age for girls and 12 and 18 years of age for boys.

This paper presents the results for a cohort of children who have now reached adulthood. Our objective when performing an SDR is to optimize gait for the whole of life, so our primary aim in this study was to investigate whether the good short term outcomes were maintained to adulthood.

METHODS

17 patients were recruited from the Oswestry database of children treated by means of an SDR. This cohort was drawn from 27 consecutive cases treated between November 1996 and January 2007. Of the 10 children excluded, 9 had yet to reach the age for their final assessment and one could not be assessed pre-operatively with the 6 camera 3D system we had at the time owing to her extensive walking aids. No children were lost to follow up and all completed all three assessment time points. The group included 6 girls with an average age of 7 years 8 months (SD 1y 1m) and 11 boys with average age of 8 years 9 months (SD 1y11m) at the time of their SDRs. Six children were GMFCS level II, nine were level III and two were level IV.

All of the patients met our selection criteria (Table 1) and had spastic cerebral palsy; two had quadriplegia and the remaining fifteen had diplegia. All the families gave informed consent for their children's data to be used for research and subsequent publication. Each child underwent an extensive pre-operative assessment including consultations with a paediatric neurologist, orthopaedic surgeon and physiotherapist, x-rays of the spine and hips and MRIs of the brain and spinal cord, full clinical examination and 3D gait analysis.

All children were treated with an open procedure. Laminotomies were performed and rootlets at the levels L1-S1 were selected for transection. The percentage of rootlets cut at each level was determined by severity of spasticity and informed by intraoperative neurophysiological monitoring. On the basis of the 13 patients for whom information is available, the mean number of rootlets cut was 33.7% (sd 7.4, range 23.1 to 50.3). All children underwent a lengthy period of in-patient rehabilitation (6-12 weeks) and were managed post-operatively in tuned rigid AFOs. They were discharged with a home exercise programme.

After SDR children were followed up regularly and orthopaedic surgery was carried out as required. No fixed protocol was used to determine when to intervene, however feet which became uncomfortable were stabilized, persistent transverse plane deformities were corrected and significant contractures resulting from residual spasticity or weakness were addressed.

In this paper gait assessment and clinical examination results are given for three specific time points: pre-SDR surgery (PRE); post-SDR surgery pre-adolescence (POST1); and post-SDR surgery post-adolescence (POST2). The target ages for POST1 and POST2 were 12yrs and 18yrs respectively for males and 10yrs and 16yrs respectively for females.

At each assessment a full clinical examination, comprising range of movements and MRC joint powers, was performed. 3D gait analyses were conducted using a Vicon motion analysis system and standard marker set (Plug-in-Gait with knee alignment devices). System specifications did vary over the study period, with between 6 and 12 cameras used and either Kistler or AMTI forceplates. Data were processed using Vicon Clinical Manager or Plug-in-Gait. At each hardware/software upgrade tests were made to confirm that there was no impact on the results of analyses. At each assessment multiple trials were collected, walking barefoot, with or without walking aids. For each patient a minimum of 5 trials was inspected for consistency and then an average was created from which key values were extracted and Gait Profile Scores (GPS) were calculated. Statistical analysis was performed using SPSS.

Clinical examination and gait data were compared across the three time points: PRE, POST1 and POST2. Random missing data were accounted for using multiple imputation methods

(16). Within subject analysis of variance was determined using Friedman and Wilcoxon signed ranks tests at a significance level of p=0.05 (2-tailed).

RESULTS

Pre-operative assessments were performed at an average of 7 months preoperatively for both boys and girls. The girls' postoperative assessments took place at 10y5m (SD 0y6m) and 16y2m (SD 0y6m). The equivalent time points for the boys were 12y5m (SD 1y2m) and 18y4m (SD 0y6m).

The additional surgical treatments (including Botulinum toxin injections) received by each child after their SDR but before the final assessment (POST2) are listed in Table 2. Conservative care such as physiotherapy or orthotic management is not recorded. Only procedures recorded in the local hospital notes are included however, as the children have been under the continuous care of the Oswestry service since their SDRs, we are confident that they have not received surgical treatment elsewhere.

Seven of the seventeen children needed no further surgical intervention following their rhizotomy. Bony procedures feature most commonly on the list, however there is some evidence of residual spasticity and contracture (nine children received Botulinum toxin injections and three had soft tissue procedures). No child went on to full single event multilevel surgery.

Eleven children maintained their GMFCS level from their pre-operative status (PRE) to maturity (POST2). Four children improved by one grade and one improved by two grades.

More detailed results of the clinical examination and gait data are given in Table 3. On examination patients have improved ranges of motion, particularly at the hip and a reduction in extensor lag at the knee was observed. Their gait patterns show better knee flexion in swing and more extension at initial contact, along with increased dorsiflexion (though a whole foot model was used so this latter finding may be influenced by flexion in the midfoot). Patients are also walking faster, taking longer steps (even allowing for height gain) and have an improved GPS. The only detrimental change observed was a small increase in anterior pelvic tilt. There were no statistically significant changes to the BMI centiles.

MRC muscle grades were collected at each assessment point, however concerns about their reliability mean they have not been included in the formal analysis in Table 2. An inspection of data from 8 key muscle groups, however, revealed that 82% of MRC grades were the same or greater postoperatively (POST 2), with 87% scoring 4 or above, compared with 79% pre-operatively.

On clinical examination we found no cases of scoliosis though we did not perform routine imaging of the spine post-operatively. One patient developed an asymptomatic spondylosis, which was detected on a lateral spinal radiograph at skeletal maturity.

The changes which have taken place over the adolescent growth spurt itself (between POST1 and POST2) are recorded in Table 4. Overall very little change has taken place. The hamstrings have become marginally less tight (popliteal angle reduced by 6 degrees) and the gastrocnemius muscle tightened by a similar amount. The patients' gait patterns have remained remarkably stable, with some additional improvement in swing phase knee flexion rates.

DISCUSSION

This paper presents the results for 17 children treated by means of SDR who have now reached adulthood. Whilst the group composition is not identical to that in our earlier paper (6) it is interesting that the overall results are comparable at 1 year follow up to those at

adulthood and the long and short term follow up results present a similar picture. There are a number of statistically significant improvements observed at adulthood of sufficient magnitude to be clinically useful. The change in GPS is, for example, double the minimally clinically important difference of 1.6 proposed by Baker et al (2012) (17). The spasticity reduction effect of SDR is particularly marked at the knee where swing phase knee flexion is smoother, improving in both velocity and amplitude. The only positive change reported at 12 months which appears to have been lost is the improved maximum knee extension in stance phase, despite maintained improvement at initial contact. In common with other centres, we observe a deterioration (increase) in anterior pelvic tilt. Overall, however, our results support the long term outcome studies which suggest that, after initial improvement, patients treated with SDR reach a functional plateau which is maintained.

We have previously reported weight increases in children following selective dorsal rhizotomy (18), particularly in those with low pre-operative walking speeds. This long term study shows no significant differences in BMI centiles between pre-operative assessment and follow up at adulthood. It should be noted, however, that one child with a very slow walking speed and large weight gain was excluded due to the absence of pre-operative 3D gait data, for reasons explained above.

The key question is the extent to which the gait pattern in adulthood can be attributed directly to the SDR performed. Children in this study were managed not only by means of rhizotomy, but also with intensive post-operative in-patient physiotherapy and careful orthotic management in tuned ankle foot orthoses. 59% also went on to have some form of orthopaedic surgery, though only three children needed any surgical correction of soft tissue contractures. Correction of long bone deformity will have made a contribution to the transverse plane gait data and surgical management of the foot will also have had an impact on sagittal plane kinematics. Both of these effects will have influenced the changes in GPS. The final result for the patients reported here will be influenced by all the interventions received. The overall rate of surgery post SDR reported here is a little lower than that typically described in the literature. Steinbok 2007 (19) in his review article quotes an average rate of around 65%. Our overall judgment is in line with his conclusion that, without SDR, all the children in this group would have gone on to receive multilevel orthopaedic surgery, though this is a speculative judgment and not possible to prove definitively without a carefully matched control group.

Other than the studies summarized in the published meta-analysis by McLaughlin et al (2002) (5) we are not aware of any randomized controlled studies of SDR and none giving long term follow up results including gait analysis data. A number of studies have attempted to create a control group to compare with their SDR patients, either from their data archive (20), from active recruitment (21), from a group of similar patients but with a slightly different presentation (22) or by allowing parental choice (23). All these approaches have clear limitations when compared to a randomized controlled trial. Given the very tight selection criteria employed, the long term follow up and the specifically timed assessments we were not able to adopt these approaches in this study.

There is, however, evidence in the literature to suggest the changes which might be expected in children with cerebral palsy as they grow. The predicted trajectory of children's GMFM scores are well defined (24). Children with GMFCS levels I and II tend to achieve a plateau, whereas those levels III-V achieve a functional peak at between 6 and 8 years of age, and after that deteriorate. The GMFM captures the functional aspects of gait but not the overall quality of the movement, as it is not designed to assess specific joint movements.

Evidence for deterioration is even stronger when measurements are taken using gait analysis, the children have been identified as surgical candidates, and the surgery has not been performed (25, 26). Gough et al 2004 (26) contend that when comparisons are made between pre-operative and post-operative data, in the absence of a control group, then positive changes are likely to be underestimated. The evidence in the literature supports our own clinical experience, which is that children with cerebral palsy such as those in our SDR programme, experience deteriorations in their gait patterns if left untreated.

In summary it is difficult to envisage a long term study of this kind being carried out as a randomized controlled trial. The literature suggests that the lack of an untreated control group is unlikely to have exaggerated the changes observed and that the improvements seen can be considered to have resulted from the overall long term management strategy employed. Selective dorsal rhizotomy was a key part of this strategy, however the additional management, including orthopaedic surgery where performed, will also have contributed to the final outcome.

CONCLUSIONS

The patients described in this paper have seen significant improvements in their ranges of motion and gait patterns from their pre-operative analysis to adulthood, an average period of 8y6m for girls and 9y5m for boys. Over this period they have received a number of interventions, in addition to SDR. Our previously published results (6) show the immediate, positive impact of the SDR when combined with rigorous selection and careful post-operative management with appropriate physiotherapy and orthotics. This current study demonstrates that the patients have navigated the adolescent growth spurt successfully and maintained significant improvements into adulthood. None of the children required a full multilevel orthopaedic surgical package of bony and soft tissue procedures at multiple joints. The evidence presented here supports the use of SDR in very carefully selected cases to provide a durable improvement in gait into adulthood.

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From History	From Examination	From Investigation
Age range 5–10 Absence of chronic conditions, eg BPD, refractory epilepsy, severe visual impairment, scoliosis Cognitive ability – IQ 70 or above Well motivated, emotionally robust child No previous multilevel surgery Good family/social support	Diagnosis spastic diplegia, quadriplegia, severe hemiplegia, HSP Spasticity moderate to severe Mean lower limb power >3 on MRC scale Movement control at least moderate Balance at least moderate Absence of severe fixed joint deformity No involuntary movements or dystonia	No hip dysplasia No basal ganglia change on MRI Weight not disproportionately greater than height
BPD, broncho-pulmonary dysplasia; HSP, hereditary spastic paraparesis.		

Table 1 Criteria used to select patients for selective dorsal rhizotomy (adapted from Cole et al., 2007)

Patient Number	GMFCS Level (PRE)	No procedures recorded	Botulinum injections	Femoral Derotation	Tibial Derotation	Muscle/Tendon Surgery	Other
1	2	X					
2	4		Bilateral Calves (3yr1)			Unilateral Streyer release (4yr5)	Unilateral subtalar arthrodesis (4yr5) Chiari pelvic osteotomy and varus femoral derotation (6yr5)
3	3				Bilateral (1yr6)		
4	2		Bilateral Calves (0yr1, 2yr0)				
5	3		Bilateral Hamstrings (1yr10) Unilateral Calf (1yr10, 2yr10, 3yr9)	Bilateral (1yr10)	Unilateral (1yr10)		
6	3		Bilateral Hamstrings (7yr1, 7yr10) Unilateral Calf (1yr10) Bilateral Calf (7yr1)	Bilateral (3yr3)			
7	2		Bilateral Hamstrings (11yr2)	Unilateral (2yr10)	Unilateral (2yr10)		Unilateral distal tibial spur excision (12yr2)
8	3		Bilateral Hamstrings (2yr7) Unilateral adductor (2yr10)			Bilateral Hamstring releases (2yr10)	
9	4	X					
10	2	X					
11	3	X					
12	2	X					
13	3	X					
14	3		Unilateral Calf (4yr2)	Bilateral (7yr4)			Unilateral foot surgery (7yr4)
15	2			Bilateral (6yr10)	Unilateral (6yr10)		
16	3		Unilateral Calf (2yr0)				Unilateral subtalar arthrodesis (2yr0)
17	3		Bilateral psoas (1yr4)			Bilateral percutaneous Achilles tendon lengthening (1yr4)	Bilateral foot surgery (1yr4)

Table 2: The additional procedures recorded for these children between their rhizotomy and their final post operative assessment (POST2). The timings for these procedures are given in brackets, as years and months after the SDR procedure was performed.

CLINICAL VARIABLES	Baseline Data PRE-SDR	Mean Change (SD) from PRE to POST2		
	Mean (SD)	Left (*p*-value)	Right (*p*-value)	Mean of L+R (*p*-value)
BMI Centiles	42.4(27.6) Male 51.4 (29.4) Female 45.5(27.8) Mean	-3.0 (31.0) Male (0.533) 6.5 (30.0) Female (0.600) 1.1 (30.23) Mean (0.906)		
HIP				
Hip Abduction (°)	15.3 (5.8)	4.4 (2.3) (0.096)	4.7(1.5)** (0.009)	4.6 (2.0)* (0.029)
Thomas Test (flexion contracture) (°)	16.0 (7.3)	-10.6 (3.0)** (0.004)	-10.3 (2.4)** (0.001)	-10.4 (2.7)** (0.001)
Anteversion (°)	39.8 (9.3)	-11.1 (3.1)* (0.018)	-13.6 (3.3)** (0.003)	-12.3 (3.2)** (0.004)
KNEE				
Maximum Extension (°)	-5.9 (6.7)	-5.4 (3.2) (0.135)	-3.4 (3.8) (0.501)	-4.4 (3.5) (0.221)
Knee Lag (°)	19.7 (11.8)	-6.1 (4.0)* (0.038)	-11.3 (3.7)** (0.003)	-8.7 (3.9)** (0.005)
Popliteal Angle (°)	51.0 (7.6)	-2.4 (3.8) (0.570)	-2.7 (3.9) (0.446)	-2.5 (3.8) (0.381)
ANKLE				
Dorsiflexion (knee bent) (°)	5.9 (9.5)	4.7 (2.9) (0.061)	6.1 (3.4)* (0.031)	5.4 (3.1)* (0.013)
Dorsiflexion (knee straight) (°)	-3.0 (7.6)	2.5 (2.8) (0.213)	3.5 (3.1) (0.214)	3.0 (3.0) (0.154)
GAIT VARIABLES				
Mean Pelvic Tilt (°)	15.0 (5.6)	5.5 (2.7)* (0.044)		
Max Stance Hip Extension (°)	-5.4 (7.9)	-4.8 (3.6) (0.435)	-0.1 (3.0) (0.407)	-2.4 (3.3) (0.723)
Knee Flexion @ Initial Contact (°)	40.3 (7.1)	-7.8 (3.7)* (0.013)	-9.7 (4.1)* (0.017)	-8.7 (3.9)** (0.009)
Max Stance Knee Extension (°)	-23.7 (10.1)	0.6 (4.6) (0.463)	4.7 (5.4) (0.084)	2.6 (5.0) (0.148)
Max Knee Flexion in Swing (°)	54.6 (7.5)	10.5 (3.8)** (0.002)	11.1 (3.2)** (0.001)	10.8 (3.6)** (0.001)
Max Rate of Knee Flexion in Swing (°/s)	129.1 (41.7)	111.1 (26.1)** (<0.001)	99.5 (21.8)** (0.001)	105.3 (23.7)** (<0.001)
Max Stance Dorsiflexion (°)	-2.2 (20.7)	12.9 (4.3)** (0.001)	20.7 (6.4)** (0.001)	16.8 (5.5)** (0.001)
Normalised Step Length (% height)	24.3 (12.8)	11.1 (3.4)** (0.002)	9.1 (3.4)** (0.006)	10.1 (3.4)** (0.002)
Walking speed (m/s)	0.77 (0.29)	0.24 (0.1)** (0.002)		
GPS	14.7 (3.6)	-3.2 (1.0)* (0.049)		

= significant at $p<0.05$

** = significant @ $p<0.001$

Table 3: This table presents the baseline values at the preoperative assessment (PRE) and the change between that time point and maturity (POST2). The left hand column gives the pre-operative values and the remaining three columns show the change. The right and left legs have been handled separately, to prevent related samples being included in the statistical analysis. The mean of left and right data is also presented as a single average figure representing each child.

CLINICAL VARIABLES	Mean Change (SD) from POST1 to POST2		
	Left (*p*-value)	Right (*p*-value)	Mean of L+R (*p*-value)
BMI Centiles	-7.3 (27.0)(0.424) Male 1.9 (31.0)(0.462) Female -4.0 (27.9)(0.463) Mea		
HIP			
Hip Abduction (°)	0.2 (2.3) (0.871)	-2.4 (1.7) (0.281)	-1.1 (2.0) (0.548)
Thomas Test (flexion contracture) (°)	-2.6 (2.9) (0.522)	-3.8 (2.6) (0.216)	-3.2 (2.7) (0.329)
Anteversion (°)	-0.8 (3.4) (0.567)	-1.4 (2.9) (0.474)	-1.1 (3.1) (0.307)
KNEE			
Maximum Extension (°)	-7.6 (3.5)* (0.037)	-2.7 (3.9) (0.635)	-5.1 (3.7) (0.191)
Knee Lag (°)	1.0 (3.92) (0.825)	-1.0 (3.25) (0.637)	0.0 (3.5) (1.00)
Popliteal Angle (°)	-6.1 (4.3)* (0.042)	-6.2 (4.1)* (0.032)	-6.2 (4.1)* (0.019)
ANKLE			
Dorsiflexion (knee bent) (°)	-3.0 (2.5) (0.269)	-5.3 (3.3)* (0.034)	4.2 (2.9) (0.066)
Dorsiflexion (knee straight) (°)	-6.0 (2.7)* (0.024)	-7.1 (3.3)* (0.013)	-6.6 (3.0)* (0.013)
GAIT VARIABLES			
Mean Pelvic Tilt (°)	-0.9 (2.8) (0.523)		
Max Stance Hip Extension (°)	-0.8 (3.9) (0.795)	1.8 (3.9) (0.523)	0.5 (3.8) (0.586)
Knee Flexion @ Initial Contact (°)	-3.6 (3.8)* (0.028)	0.6 (4.8) (0.435)	-1.5 (4.3) (0.098)
Max Stance Knee Extension (°)	-1.6 (5.0) (0.687)	-0.6 (6.2) (0.309)	-1.1 (5.6) (0.332)
Max Knee Flexion in Swing (°)	4.3 (4.0) (0.740)	4.2 (3.2) (0.309)	4.2 (3.7) (0.722)
Max Rate of Knee Flexion in Swing (°/s)	44.0 (31.7)* (0.033)	22.2 (27.9) (0.170)	33.1 (29.5)* (0.044)
Max Stance Dorsiflexion (°)	-0.7 (3.4) (1.00)	-0.8 (2.8) (0.523)	-0.7 (3.1) (0.723)
Normalised Step Length (% height)	2.3 (2.7) (0.943)	1.43 (3.0) (0.943)	1.9 (2.8) (0.813)
Walking speed (m/s)	0.01 (0.1) (0.619)		
GPS	0.3 (1.1) (0.301)		

* = significant at $p<0.05$

** = significant @ $p<0.001$

Table 4: This table presents the change from the pre-adolescent post-operative assessment (POST1) to maturity (POST2). It therefore represents the change which took place over the adolescent growth spurt (in girls from age 10 to 16 and in boys from age 12 to 18). As in Table 3 the right and left legs have been handled separately and the mean of left and right data is also presented.