Selective dorsal rhizotomy: meta-analysis of three randomized controlled trials

Authors' objectives
To identify the factors responsible for the different outcomes of the studies, and to clarify whether selective dorsal rhizotomy (SDR) makes a statistically-significant contribution to the functional improvement of children with spastic cerebral palsy up to one year after the operation.

Searching
The review's authors had been involved in three relevant RCTs, and undertook a literature search to find out whether any other trials had been conducted as of December 2000. This included a search of MEDLINE, the Cochrane Controlled Trials Register and abstracts from recent scientific meetings, and contact with other researchers in the field.

Study selection
Study designs of evaluations included in the review
Randomised controlled trials (RCTs) were included. Randomisation was undertaken using a block age of less than 6 years and of at least 6 years; a block age of 3 to 11, 12 to 18 years and ambulatory status; or using no blocking. The duration of follow-up for the included studies was 9 months, one year, and one to 2 years. Two included trials were single-blind studies, while a further study was not blind.

Specific interventions included in the review
SDR. The included studies compared SDR plus physiotherapy (PT) with or without occupational therapy versus PT with or without occupational therapy. PT consisted of programmes of stretching, strengthening, and training in functional movements intended to enhance mobility.

Participants included in the review
Children with spastic cerebral palsy. Children with dystonia, athetosis, ataxia or severe visual impairment were excluded. The mean age of the children in the included studies was 5.5 years (standard deviation, SD=2.8) and the percentage of females was 47%. The mean baseline score using the lower extremity Ashworth scale (see Other Publications of Related Interest no.1.) was 2.9 (SD=0.8), the mean baseline total Gross Motor Function Measure (GMFM) (see Other Publications of Related Interest no.2) was 2.9 (SD=0.8), and the mean baseline GMFM-66 was 62.5 (SD=16.0).

Outcomes assessed in the review
The Ashworth scale was used to measure spasticity, while the GMFM and GMFM-66 (an improved scaling method to that of the GMFM) were used to measure function. The primary outcome variables used in the included studies were GMFM total and diplegia composite, and spasticity, measured using the Ashworth scale with or without myometry or electromechanical torque measurement.

How were decisions on the relevance of primary studies made?
The authors do not state how the papers were selected for the review, or how many of the reviewers performed the selection. However, the review's authors had been involved in the included studies.

Assessment of study quality
The authors do not state that they assessed validity. However, the review's authors were involved in the three included studies, and details of randomisation and blinding were reported in summary tables.

Data extraction
The authors do not state how the data were extracted for the review, or how many of the reviewers performed the data extraction.
extraction.

The extracted data included: treatment; randomisation details; blinding; duration of follow-up; inclusion criteria; criteria for dorsal root tissue transection; post-operative management; time of assessment; primary outcome variables; secondary outcome variables; difference to detect; and the data analysis used in the primary outcome papers. Complete data were obtained for all the variables reported with the exception of baseline ambulatory status. No new data were collected for the review.

The markers of severity used by the studies were initial diagnosis (spastic diplegia), qualitative descriptors (mild, moderate or severe), ambulatory status and baseline GMFM score. For the review, the Gross Motor Function Classification System (GMFCS) was used to provide a means of rank-ordering the functional severity of cerebral palsy on age-adjusted clinical descriptors. A developmental paediatrician familiar with the GMFCS, and masked with respect to participant identification and study group assignment but not study site, retrospectively reviewed the selected clinical descriptions of all the participants. Data from each child were assigned a GMFCS baseline score, which was entered into the multivariate analysis.

Methods of synthesis
How were the studies combined?
For each outcome, the change in SDR plus PT group was compared with that in the PT-only group using two methods: blocked Wilcoxon's test (blocking on site; see Other Publications of Related Interest no.3); and analysis of variance, including factors for treatment group, site, and a treatment by site interaction. Separate multiple regression analyses were conducted to evaluate the effect of characteristics or the effect of SDR on outcome, as measured by the Ashworth, GMFM and GMFM-66 scales. These analyses used backward selection and the following additional predictors: age, gender, birth weight, ambulatory status, baseline GMFM-66, and baseline lower-extremity Ashworth score. Site was included in the models, regardless of significance; all other variables required a significance level of p less than 0.05 to be retained. Once the significant main effects were identified, two-way interactions among the included variables were evaluated.

How were differences between studies investigated?
The differences were described in a narrative summary. Characteristics of the three samples were also compared using a Kruskal-Wallis distribution-free analysis of variance for continuous or ordered variables, and chi-squared or Fisher's exact tests for unordered categorical variables.

Results of the review
Three RCTs involving 90 children were included. Of these, 82 were aged under 8 years old and 65 had GMFCS level II or III disability.

Two studies showed a statistically-significant advantage in the functional outcome for SDR compared with PT alone, while the third study showed no advantage. This third study was the only one to include children (n=8) who were at least 8 years of age at baseline; the study sample was also slightly more heterogeneous with respect to intellectual function and cause of cerebral palsy.

The pooled Ashworth data analysis confirmed a reduction of spasticity with SDR plus PT (mean change score difference -1.2; Wilcoxon p<0.001). The pooled GMFM data revealed greater functional improvement with SDR plus PT (difference in change scores 4.0; Wilcoxon p=0.008). When the GMF-66 scores were used, the mean difference in change score for the pooled data was smaller (2.6) and remained statistically significant (Wilcoxon p=0.002). No baseline characteristics were found to be significantly associated with outcome in the multivariate analysis. Multiple regression in the SDR plus PT group revealed a direct relationship between the percentage of dorsal root tissue transected and functional improvement: patients who had a larger amount of dorsal root tissue transected had more improvement on GMFM (p<0.001), GMFM-66 (p=0.02), and Ashworth score (p=0.03).

The potential effects of outliers and of sectioning S2 root tissue were examined by eliminating relevant data from the analyses. No appreciable effects were identified.
Authors' conclusions
SDR plus PT was efficacious in reducing spasticity in children with spastic diplegia, and it had a small positive effect on gross motor function. There appeared to be a direct relationship between the percent of dorsal root tissue transected and the magnitude of the gain in function, although this statistical correlation may not reflect a true cause effect relationship.

CRD commentary
This was a meta-analysis of individual patient data, which was of reasonable quality. The aims of the review were clearly stated and details of the inclusion criteria were reported. The authors had been involved in three included RCTs, and they undertook a literature search to identify any further trials conducted as of December 2000. Only three electronic databases were searched and details of the search strategy were not reported. The possibility of publication bias was not discussed. Relevant details of the primary studies were presented in tabular format. No information was reported on the comparability of the characteristics of the two treatment groups. The authors did not state whether their meta-analysis was conducted on an intention to treat basis. Two participants were lost to follow-up in one trial and five in another. For this second trial, initial analyses were reported to have been undertaken using intention to treat. Differences between the included studies were assessed in both a narrative summary analysis and a statistical analysis.

The studies were pooled appropriately and the authors' conclusions follow from the results. A detailed investigation was made of the possible factors associated with variations in the results of different trials, although the results were only partially presented.

Implications of the review for practice and research
Practice: Based on both clinical experience and the review data, the authors speculate that SDR might be most effective for a child between 3 and 18 years of age whose functional level falls into GMFCS III and IV. Children in this age range are easy to assess, tolerate the physiotherapy regimens, and have academic and social demands that can accommodate an intensive intervention. Children with more severe cerebral palsy may have more potential gain from an invasive procedure.

Research: The authors state that further sizable well-studied cohort studies of individuals with spastic diplegia followed to adulthood are required, to assess the beneficial effect of surgical treatment in terms of the long-term outcomes.

Funding
United Cerebral Palsy Research and Educational Foundation; National Institute for Neurological Disease and Stroke, grant number R01- NS27867; British Columbia Health Care Research Foundation; Easter Seal Research Institute of Canada; United Cerebral Palsy Research and Educational Foundation.

Bibliographic details

PubMedID
11811645

Other publications of related interest
Indexing Status
Subject indexing assigned by NLM

MeSH
Cerebral Palsy /surgery; Child; Child, Preschool; Disabled Children; Female; Ganglia, Spinal /surgery; Humans; Male; Motor Skills; Randomized Controlled Trials as Topic; Rhizotomy; Treatment Outcome

AccessionNumber
12002000262

Date bibliographic record published
28/02/2003

Date abstract record published
28/02/2003

Record Status
This is a critical abstract of a systematic review that meets the criteria for inclusion on DARE. Each critical abstract contains a brief summary of the review methods, results and conclusions followed by a detailed critical assessment on the reliability of the review and the conclusions drawn.