Spasticity in children and young people with non-progressive brain disorders: management of spasticity, co-existing motor disorders and their early musculoskeletal complications

Orthopaedic surgery

Bibliographic details	Number of Participants Characteristics	Intervention characteristics	Outcome measures and results	Quality assessment	Reviewer comment
Periodical Archives of Physical Medicine and Rehabilitation **Authors** Yang,E.J., Rha,D., Kim,H.W., Park,E.S. **Year of publication** 2008 **Study location** South Korea **Ref ID** 111548 **Type of study** Retrospective cohort study **Aim of study** To compare the effects of BoNT-A injection into the hip adductor muscles with soft tissue surgery on hip displacement and identify the factors affecting outcomes of both BoNT injection and soft tissue surgery	**Inclusion Criteria** Children with CP admitted to hospital between Feb 2004 and Mar 2007 1) who had bilateral spastic CP 2) whose first hip radiographs were taken under 6 years of age 3) in whom radiographs of the hips were taken at least 3 times in intervals of more than 6 months. **Exclusion Criteria** Children with both a soft-tissue surgery and BoNT-A injection during the follow-up period were excluded **Baseline characteristics** 194 children with spastic CP were enrolled Diplegia : n=116, Quadriplegia : n=78 High functioning group : GMFCS I and II n= 58 Low functioning group : GMFCS III, IV and V n= 136	No intervention (138 hips of 69 children) Soft tissue surgery (130 hips of 65 children) Soft tissue surgery of hip adductor muscles BoNT-A (120 hips of 60 children) BoNT-A injection into hip adductor muscle BoNT-A brand: Not stated Average dose : 3U/kg body weight standardised by body weight during this time period Injection details : 1 ml syringe with 27-G needle Solution : contents of one vial of BoNT-A dissolved in 2ml isotonic saline Guidance : ultrasonography 7 children received an additional BoNT-A injection into both hip adductor muscles during the follow up period.	Hip Migration Percentage (MP) MP was measured by calculating the % femoral head lying outside the lateral border of the acetabulum as defined by bony landmarks on an anteroposterior pelvis radiograph Mean change in hip migration percentage (%) No intervention group : 4.7±10.3 BoNT group : -1.6±8.4 Surgery group : -3.3±6.1 Mean change per year in hip migration percentage (%) No intervention group : 4.4±11.3 BoNT group : -0.7±6.5 Surgery group : -1.6±4.4 Mean change per year in hip migration percentage (%) - high functioning children No intervention group (n=68) :	Study type : retrospective cohort study reviewing case notes Allocation to treatment unrelated to confounders : Unclear Attempt to balance groups for confounders : Yes Groups comparable at baseline : Yes (except for male: female ratio = 85%/15% in surgical group) Participants received similar care (except intervention?) : Yes Participants blinded to treatment : No Caregivers blinded to treatment : No No of participants for whom no data was available (each treatment arm) : None Length of follow up appropriate : Yes (mean 22 months) Definitions of outcomes given : Yes Outcomes assessed with valid	Ethical approval : Not stated Consent : Not stated Funding : Not stated

	Groups according to severity of hip displacement at initial MP assessment Mild subluxated group 20%≤MP<40% n=120 Moderate subluxated group 40%≤MP<60% n=70 Severe subluxated group 60%≤MP<90% n=4 Mean age at initial radiograph 39.3±12.9 months (range 18 to 70 months) Mean age at final radiograph 62.0±17.7 months (range 37 to 174) Mean duration of follow up 22.9±11.8 months(range 18 to 108) No significant differences at baseline between no intervention, soft tissue surgery and BoNT-A groups for any of the following : GMFCS score, initial MP, initial age, final age and duration of follow up, proportion of high and low functioning participants, proportion of participants with mild, moderate or severe subluxation		-2.8±5.0 BoNT group (n=40 legs) : -2.4±5.2 Surgery group (n=28 legs) : -3.4±4.8 Mean change per year in hip migration percentage (%) - low functioning children No intervention group (n=182 legs) : -0.5±5.6 BoNT group (n=90 legs) : -0.0±6.9 Surgery group (n=72 legs): -1.0±4.1 For each intervention (no invervention, BoNT and surgery) the higher functioning group's Mean Change HM% per year was statistically significantly greater than the low functioning group.	method : Yes Similar length of follow up for different groups: Yes Similar number of participants completed tx in each group : Yes Investigators blinded to patients' exposure to intervention : Unclear Investigators blinded to impt confounders/prognostic factors : Unclear Outcome assessors blinded to treatment :Unclear	

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Periodical European Journal of Neurology **Authors** Molenaers,G., Desloovere,K., De,Cat J., Jonkers,I., De,Borre L., Pauwels,P., Nijs,J., Fabry,G., De,Cock P. **Year of publication** 2001 **Study location** Belgium **Ref ID** 117421 **Type of study** Retrospective cohort study **Aim of study** To provide objective evidence of two treatment options (multilevel botulinum toxin type A and multilevel surgery) for children with cerebral palsy. To evaluate the success of two multilevel treatment strategies for children with generalised joint impairments when each are applied in normal clinical conditions.	**Inclusion Criteria** Children randomly selected from a larger cohort of children treated between 1998 and 1999 at University Hospital Leuven. Children with a diagnosis of spastic CP with independent barefoot walking without walking aids before and after treatment. **Exclusion Criteria** None stated **Baseline characteristics** BoNT and casting group N= 29 pts, 43 treated limbs Diagnosis = 14 diplegia, 15 hemiplegia Age mean (range) = 6 years 2 months (4yrs 3m to 9 yrs 10m) Post treatment evaluation = 2 months post treatment Orthosis use pretreatment : Daytime - 5pts used leafspring AFOs, 6 pts used hinged AFOs, 1 pt used fixed AFOs. Night - 5 patients (3 limited use) Orthosis use posttreatment : Daytime - 19 pt used leafspring AFOs, 7 pts used hinged AFOs. Night - 26patients (5 limited use) Therapy pre-treatment = Mean of 2.4 sessions/wk Therapy post-treatment = Mean of 2.9 sessions/wk	BoNT and casting treatment BoNT A type : Botox Dilution : 50U/ml Maximum total dose : 50U Botox -A per site Dosage and Muscle Selection : Total dose averaged 25.5U/body weight (range 20-31 U/kg BW) for children with diplegia and 13.7U/body weight (range 6-20U/kg BW) for children with hemiplegia. Injections were fine tuned on a patient by patient basis following objective examination using full gait analysis and an extended clinical examination. Between 2 and 5 muscles were injected in each treated limb in one session. All patients received injections in gastrocnemius and medial hamstrings. Other muscles injected included soleus, tibialis posterior, adductors and iliopsoas Sedation and pain management : child sedated with mask anaesthesia Casting All patients were casted at the distal joints immediately before or after injections to	Outcomes were assessed at 2 months in the BoNT group and 12 months in the surgery group as it was decided to evaluate the children at a point of (presumed) maximum effect of treatment. BoNT group n=29 patients, 43 limbs Surgery group n=23 patients, 43 limbs Mean walking speed m/s BoNT group : Pre treatment = 1.06 (0.2) Post treatment = 1.03 (0.2) Surgery group : Pre treatment = 0.9 (0.2) Post treatment = 0.8 (0.2)	Study type : retrospective cohort study Allocation to treatment unrelated to confounders : Unclear Attempt to balance groups for confounders : No Groups comparable at baseline : No, Proportion of diplegia to hemiplegia different in each group, Age - BoNT group younger than the Surgery group. "Previous BoNT" higher in BoNT group compared to Surgery group (9 pts vs 1pt) and Previous Surgery" higher in Surgery group compared to BoNT group (11 pts vs 1pt) Participants received similar care (except intervention) : Unclear, description suggests that the surgical group may have received more intensive post-intervention therapy Participants blinded to treatment : No Caregivers blinded to treatment : No No of participants for whom no data was available (each treatment arm) : None Length of follow up appropriate : Assessments made at time of presumed maximum efficacy ie 2 months post treatment follow up in	Ethical approval : Not stated Consent : Not stated Funding : Not stated

	Previous surgery = 1 patient Previous BoNT treatment = 9 patients <u>Surgery group</u> N= 23 patients, 43 treated limbs Diagnosis = 20 diplegia, 3 hemiplegia Age (mean) (range) = 13 yrs 5 months (7yrs 4m to 21yrs 7m) Post treatment evaluation = 12 months post treatment Orthosis use pretreatment : Daytime - 1pt used leafspring AFOs, 4 pts used hinged AFOs. Night - 1 patient (limited use) Orthosis use posttreatment : Daytime - 5 pts used leafspring AFOs, 3 pts used hinged AFOs, 2 pts used ground reaction AFOs. Night - 18 6patients (1 limited use) Therapy pre-treatment = Mean of 2.6 sessions/wk Therapy post-treatment = Mean of 3.6 sessions/wk Previous surgery = 11 patients Previous BoNT treatment = 1 patient	correct mild contractures and to enhance the effect of the injections. Serial stretching casts (for a period of 10-28 days) were applied to bothe lower limbs (for children with diplegia and hemiplegia) with the ankle joint in neutral position or in 5° of dorsiflexion and the subtalar joint and midtarsal joints in a neutral position. On average cases were reapplied every 12 days. <u>Surgery</u> 3D gait analysis was used to delineate the gait deviations of each patient and to help to plan the surgical intervention. 7 patients had soft tissue surgery only, 16 patients had soft tissue surgery combined with corrections of bony deformities. Soft tissue procedures included: - Lengthening of the psoas, adductor longus, and medial hamstrings - Rectus femoris transfer to either gracilis or semitendinosus - Procedures involving gastrocnemius (Stryer or Achilles tendon lengthening in children with hemiplegia - Lengthening of peroneus		BoNT group and 12 months post treatment follow up in surgery group Definitions of outcomes given : Yes, outcomes assessed as part of gait analysis (details of instruments used given) Outcomes assessed with valid method : Yes Similar length of follow up for different groups: No 2 months post treatment follow up in BoNT group and 12 months post treatment follow up in surgery group Similar number of participants completed tx in each group : Yes Investigators blinded to patients' exposure to intervention : No Investigators blinded to impt confounders/prognostic factors : No Outcome assessors blinded to treatment : No	

		- Tibialis posterior lengthening or transfer - Tibialis anterior transfer - Flexor hallucis lengthening Bony deformity corrections included: - Acetabular corrections - Proximal femoral varus derotation osteotomy - Tibial realignment and foot stabilisation surgery (calcaneus lengthening combined with medial soft-tissue shortening and subtalar arthrodesis) All patients received a combination of surgical procedures at 3 levels in one session. All patients after BoNT or surgery had appropriate physiotherapy and orthotic management involving day orthoses and night splinting. This is described as "intensive" rehabilitation following surgery			

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Periodical Journal of Pediatric Orthopedics **Authors** Gorton,G.E.,III, Abel,M.F., Oeffinger,D.J., Bagley,A., Rogers,S.P., Damiano,D., Romness,M., Tylkowski,C. **Year of publication** 2009 **Study location** USA **Ref ID** 100823 **Type of study** Prospective cohort study **Aim of study** To prospectively examine whether lower extremity musculotendinous surgery in ambulatory children with CP improves impairments and function measured by gait and clinical outcome tools beyond changes fouond in a concurrent matched control group	**Inclusion Criteria** Diagnosis of CP, GMFCS level I to III, age 4 to 18 years, ability to complete gait analysis. This study of ambulatory children with CP is part of a 6 year prospective multicentre study across 7 paediatric orthopaedic facilities. **Exclusion Criteria** Earlier SDR, orthopaedic surgery within the previous year, BoNT injectinos within the last 6 months or a currently operating baclofen pump **Baseline characteristics** Total participants in each group Surgical Group : 75 who had lower extremity surgery and complete follow up assesment at 12 m after surgery Non-surgical Group : 75 who did not have surgery, either because is was not recommended based on full clinical assessment including 3D gait analysis or because the family did not elect to move forward with surgery during the study period, and who received standard care Variables used to match surgical and nonsurgical	Surgery Procedures included both soft tissue and bony surgery Soft tissue procedure only : 50/75 Bony procedures only : 5/75 Soft tissue and bony procedures :20/75 Soft tissue procedures included: rectus femoris transfer, hamstring lengthening, heelcord lengthening, adductor lengthening, psoas lenthening and other foot/ankle transfers Bony procedures included : femoral derotation osteotomy, tibia/fibula rerotation osteotomy, lateral column lengthening Standard care Observation, stretching and strengthening exercises, bracing and medication management. No surgery, BoNT injections or ITB pump insertion.	GMFM Dimension D Baseline Surgical = 83.0 (17.9) Baseline Nonsurgical= 82.2 (18.7) Follow-up Surgical = 83.0 (1.2) Follow-up Nonsurgical= 84.6 (1.2) ANCOVA P* = 0.331 MCID (0.5) = 1.8 GMFM Dimension E Baseline Surgical = 74.5 (26.4) Baseline Nonsurgical= 73.9 (26.1) Follow-up Surgical = 73.8 (1.3) Follow-up Nonsurgical= 76.0 (1.3) ANCOVA P* = 0.192 MCID (0.5) = 2.6 GMFM-66 Baseline Surgical = 75.0 (12.7) Baseline Nonsurgical= 74.4 (12.9) Follow-up Surgical = 75.0 (0.6) Follow-up Nonsurgical= 76.2 (0.6) ANCOVA P* = 0.172 MCID (0.5) = 1.3 PedsQL Physical Functioning Baseline Surgical = 55.8 (19.8) Baseline Nonsurgical= 59.0 (19.7) Follow-up Surgical = 60.5 (2.2) Follow-up Nonsurgical= 54.7	Study type : prospective cohort study Allocation to treatment unrelated to confounders : No Attempt to balance groups for confounders : Yes Groups comparable at baseline : Yes for matching variables Participants received similar care (except intervention) : Unclear Participants blinded to treatment : No Caregivers blinded to treatment : No No of participants for whom no data was available (each treatment arm) : None Length of follow up appropriate : Yes, 1 year Definitions of outcomes given : Yes, validated tools Outcomes assessed with valid method : Yes Similar length of follow up for different groups: Yes Similar number of participants completed tx in each group : Yes Investigators blinded to patients' exposure to intervention : No Investigators blinded to impt confounders/prognostic factors : No Outcome assessors blinded to	Ethical approval : Institutional Review Boards Consent : Obtained for participants Funding : Shriner Hospitals for Children Clinical Outcomes Study Advisory Board Grant no 9140

	groups at baseline Age Surgical Group : 11.3±3.1 Non-surgical Group : 11.3± 2.9 Height Surgical Group : 139.7 ± 19 Non-surgical Group : 139.8±18.3 Weight Surgical Group : 38.7±16.5 Non-surgical Group :40.5±18.4 GMFM Dimension E (%) Surgical Group : 74.5±26.4 Non-surgical Group : 73.9±26.1 Groups were not matched on pre-operative gait kinetcs, joint spasticity or other clinical indications typically used in determining appropriateness for musculoskeletal surgery.		(2.1) ANCOVA P* = 0.039 MCID (0.5) = 12.7 PedsQL Emotional Functioning Baseline Surgical = 67.6 (17.5) Baseline Nonsurgical= 66.9 (16.0) Follow-up Surgical = 68.8 (2.0) Follow-up Nonsurgical= 64.7 (1.9) ANCOVA P* = 0.109 MCID (0.5) = 10.5 PedsQL Social Functioning Baseline Surgical = 55.1 (20.5) Baseline Nonsurgical= 56.5 (19.2) Follow-up Surgical = 59.4 (2.5) Follow-up Nonsurgical= 55.4 (2.5) ANCOVA P* = 0.221 MCID (0.5) = 12.8 PedsQL School Functioning Baseline Surgical = 64.9 (17.3) Baseline Nonsurgical= 61.8 (16.3) Follow-up Surgical = 67.1 (2.0) Follow-up Nonsurgical= 64.6 (1.9) ANCOVA P* = 0.320 MCID (0.5) = 12.3 Velocity (%normal)	treatment : No	

			Baseline Surgical = 77.8 (23.7) Baseline Nonsurgical= 78.9 (22.3) Follow-up Surgical = 79.1 (2.0) Follow-up Nonsurgical= 78.6 (1.9) ANCOVA P* = 0.844 MCID (0.5) = 9.1		

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Periodical Journal of Bone and Joint Surgery - American Volume **Authors** Thomason,P., Baker,R., Dodd,K., Taylor,N., Selber,P., Wolfe,R., Graham,H.K. **Year of publication** 2011 **Study location** Australia **Ref ID** 132766 **Type of study** Randomised controlled study **Aim of study** To evaluate the magnitude of change between groups and over time on the basis of gait indices, physical measures, function, activity, mobility and health-related quality of life following single-event multilevel surgery in children 6-12 years old who had spastic diplegia.	**Inclusion Criteria** 1) Confirmed diagnosis of cerebral palsy with registration in the Victorian Cerebral Palsy Register 2) A spastic movement disorder 3) Aged 6 -12 years 4) GMFCS level of II or III 5) Suitability for multilevel surgery **Exclusion Criteria** 1) Diagnosis of dystonia 2) Prior orthopaedic surgery, selective dorsal rhizotomy, or intrathecal baclofen therapy 3) Any reasons why delaying surgery might cause harm, such as hip migration in excess of 25% on radiographs, painful breakdown of the midfoot, and progressive crouch gait (defined as a loss of knee extension of > 10 degrees in late stance) **Baseline characteristics** N=19 Baseline characteristics are reported in an appendix, not included with the paper.	This was a randomised controlled trial comparing: - single event multi-level surgery followed by intensive postoperative physical therapy - physical therapy alone Randomisation A consecutive sample of 30 children with spastic diplegic CP were assessed for eligibility, of which 19 met the inclusion criteria and were randomised. The randomisation was performed by the trial statistician, using a minimisation approach to ensure that the groups were well-matched. Random allocation was done via a computer program. Minimisation was based on GMFCS level (I or II), age (less or older than 9 years old), and type of surgery (osseous only, soft tissue only, or both). Interventions Surgery group (n=11) Single event multilevel surgery was defined as: at least one surgical procedure performed at two different anatomical	1. Comparative data: results of between group comparisons at 12 months Walking: GPS (median (IQR)) Baseline surgical: 13.7 (11.9, 15.2) Baseline control: 14.6 (10.5, 15.8) 12-month surgical: 9.1 (8.6, 12.6) 12-month control: 15.7 (13.9, 16.2) Difference between groups in change at 12 months (95% CI): -5.5 (-7.6, -3.4) p<0.001 Walking: GGI score (mean (SD)) Baseline surgical: 353 (211) Baseline control: 370 (194) 12-month surgical: 153 (81) 12-month control: 381 (196) Difference between groups in change at 12 months (95% CI): -218 (-299, -136) p<0.001 Function: GMFM-66 (mean (SD))	Small sample size; sample size calculation was not performed, due to the lack of pilot data Study type: randomised controlled trial, with additional prospective follow-up of one arm Appropriate randomisation: yes Allocation concealment: yes Groups comparable at baseline: unclear Participants blinded: no Outcome assessors blinded: unclear Participants received similar care except for intervention: yes Number of participants for whom no data was available: None Appropriate length of follow-up: yes	No children were lost to follow-up. Only the surgical arm were followed up for 24 months, as the control arm received surgery after 12 months. Ethical approval: Yes - granted by the Ethics in Human Research Committee of the Royal Children's Hospital, Melbourne Consent: Yes - informed written consent was obtained from parents of eligible children, following a minimum of two detailed interviews with the treating surgeons and the study coordinator. Funding: Received from the Hugh Williamson Foundation, the Murdoch Children's Research Institute, and the National Health and Medical Research Council, the Centre for Clinical Research Excellence in Gait Analysis and Gait Rehabilitation. Funding for the rehabilitation program was provided by the Post Intervention Physical Therapy Program

		levels (the hip, knee or ankle) on both sides of the body. The surgical recommendation was tailored to the child's needs as determined by a comprehensive evaluation, including a standardised physical examination, radiographic evaluation, and instrumented gait analysis. The multilevel surgical program included muscle tendon lengthening, tendon transfer, rotational osteotomy, and stabilisation of the hip and foot according to published guidelines. A total of 85 procedures were performed, with a mean of 8 procedures per child (SD 4). The children allocated to the surgical group had surgery performed by two experienced surgeons, within 4 weeks of the baseline assessment. Perioperative antibiotics and epidural infusions of 0.25% bupivacaine weer used. Children remained as inpatients for 5-7 days following surgery, and were discharged wearing below-the-knee plaster casts, with knee immobilisers and the use of appropriate assistive devices, as indicated by their GMFCS level.	Baseline surgical: 65.3 (11.1) Baseline control: 70.3 (11.3) 12-month surgical: 66.1 (8.9) 12-month control: 69.8 (11.4) Difference between groups in change at 12 months (95% CI): 0.3 (-4.5, 5.0) NS <u>Quality of life: CHQ-PF50 scores (mean (SD))</u> <u>a. Physical function</u> Baseline surgical: 47 (26) Baseline control: 62 (35) 12-month surgical: 58 (26) 12-month control: 76 (25) Difference between groups in change at 12 months (95% CI): -14 (-39, 11) NS <u>b. Social/emotional</u> Baseline surgical: 69 (34) Baseline control: 89 (21) 12-month surgical: 65 (36) 12-month control: 97 (8) Difference between groups in change at 12 months (95% CI): -32 (-62, -2) p<0.05 <u>c. Family cohesion</u>		

		The surgical group were assessed at 3 and 6 weeks postoperatively to check healing and provide custom-fitted ankle-foot orthoses. Physical therapy in the first 3 months was aimed at regaining function lost as a result of surgery. This was followed by an intensive program performed 3 times a week for twelve weeks, aimed at improving range of motion, strength, balance, and function. Control group (n=8) The control group underwent a progressive resistance strength training program. They continued their routine physical therapy program for the first three month. In the second three months, they commenced the lower limb progressive resistance strength training program, which was performed three times per week for twelve weeks in their usual therapy sessions. Exercises were targeted at strengthening the hip abductors and extensors, knee extensors, and ankle plantar flexors.	Baseline surgical: 72 (20) Baseline control: 69 (20) 12-month surgical: 83 (13) 12-month control: 69 (20) Difference between groups in change at 12 months (95% CI): 14 (-2, 30) NS 2. Case series data: results of 24 month follow-up in surgery group (n=11) GPS (median (IQR) Baseline: 13.7 (11.9, 15.2) Follow-up: 9.1 (7.8, 9.6) Difference (95% CI): -5.4 (-7.5, -3.3) p<0.05 GGI score (mean (SD)) Baseline: 353 (211) Follow-up: 139 (80) Difference (95% CI): -213 (-327, -100) p<0.05 GMFM-66 score (mean (SD)) Baseline: 65.3 (11.1) Follow-up: 70.2 (10.1) Difference (95% CI): 4.9		

		The frequency, duration and cost of therapy were matched for the treatment and control groups. Outcome assessment Quantitative 3D gait data were collected using a six-camera Vicon 370 system. Reflective markers were attached to the osseous landmarks. Gait Profile Score (GPS) and Gillette Gait Index (GGI) were assessed at baseline and at 12 months postoperatively. The children in the control group exited the study after the 12-month assessment and progressed to surgery. The children who have been randomised to surgery continued to be followed in a prospective cohort study for a minimum of three years. Results from 24 months are reported. Patient reported outcomes were assessed with the use of the Child Health Questionnaire - Parent Form 50 (CHQ-PF50), Australian authorised adaptation.	(0.98, 8.7) p<0.05 Quality of life: CHQ-PF50 physical function domain (mean (SD)) Baseline: 47 (26) Follow-up: 69 (18) Difference (95% CI): 22 (4, 39) p<0.05 Adverse events related to surgery (n (%)) - Mild (spontaneously resolving): 3 (27.3) [Three children had a total of 4 mild adverse events related to poor postoperative pain management. In 2 children, this was due to postoperative epidural malfunction. One child had difficulties with pain and excessive consumption of codeine, which was followed by constipation with emesis.] - Moderate (resolved completely following simple treatment): 3 (27.3) [Two had pain over		

		Analysis - An analysis-of-covariance between the groups at 12 months and a linear regression analysis with standard errors for comparison of baseline and 24 month values within the surgical group were carried out for all outcome measures.	femoral osteotomy plates, which resolved with implant removal. One had foot pain following os calcis lengthening, which resolved by 6 months after the surgery.]		