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A1

Recurrence of lower extremity torsional deformities in ambulatory children with cerebral palsy

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Background/Objectives: Children with cerebral palsy (CP) often undergo derotational osteotomy of the femur (FDRO) and/or the tibia (TDRO) for transverse plane mal-alignment. Rotational deformity can recur after surgical correction. The purpose of this study was to examine factors contributing to recurrence of femoral and/or tibial torsion following derotation osteotomy (DRO) in ambulatory children with CP.

Design: Retrospective cohort study.

Participants and setting: 99 children with CP (GMFCS level I–IV), average age 7.7 (2.6) years; 59 males, 40 females) treated at a tertiary pediatric referral center.

Methods: We identified children with CP who underwent DRO of the femur (FDRO) and/or tibia (TDRO) and who had both pre- and post-operative gait analysis (1–11y after surgery). Retrospective review was conducted of surgical history, physical exam measures, and dynamic gait characteristics. Recurrence of rotational problems was positive if subsequent DRO was either done or recommended following postoperative gait analysis. Predictors of recurrence were evaluated using logistic regression.

Results: 160 limbs in 99 patients were included. Of these, 25 (15.6%) developed recurrent torsion of the femur and/or tibia. Predictors of overall recurrence were GMFCS level (Odds Ratio 1.84, 95% CI 1.01-3.38) and preoperative thigh-foot angle (TFA) (OR 1.04, 95% CI 1.00-1.08). For patients who underwent FDRO without concomitant TDRO, recurrence (6/69) was predicted by preoperative TFA (OR 1.54, 95% CI 0.96-2.49). When isolated external TDRO was done without concomitant FDRO, recurrence was seen in 3/32, and overcorrection was seen in 4/32 limbs. When isolated internal TDROs were performed, recurrence was seen in 2/11 limbs, with no overcorrections. There were no preoperative predictors of internal or external tibial torsion recurrence. For patients in whom concomitant FDRO and TDRO were performed, results varied according to direction of tibial DRO. When both the femur and tibia underwent external DRO, all recurrences were in the femur (5/39) and no recurrence or overcorrection was seen in the tibia. When external FDRO was combined with internal TDRO, higher rates of recurrence were seen in both bones (femur 2/7, tibia recurrence 3/7, tibia overcorrection 2/7). Higher GMFCS level (greater disability) and more external preoperative TFA were predictors of recurrence in this group. Age at surgery and length of follow-up were not predictive of recurrence.

Conclusions: Higher GMFCS level and more external preoperative TFA were predictors of recurrence of torsional deformity after DRO. Tibial DRO appears less predictable than FDRO, as both recurrence and overcorrection were seen at that level. The highest need for re-operation was seen after external FDRO combined with internal TDRO. These findings may help refine indications for DRO in this patient population.

A2

Long-term outcome of single event multilevel surgery including distal hamstring lengthening in spastic diplegia

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Background/Objectives: Distal hamstring lengthening (DHL) is a commonly performed procedure in flexed knee gait. However, the necessity of this procedure has been challenged due to the concerns on adverse effects in long-term follow-up. This retrospective study was undertaken to investigate the long-term outcome of single event multilevel surgery (SEM-LS), including bilateral DHL, in ambulatory patients with cerebral palsy using 3D gait analysis.

Design: Retrospective study.

Participants and setting: Twenty-nine ambulatory patients with spastic diplegic cerebral palsy (GMFCS level I-III) who had undergone SEMLS including bilateral DHL were included.

Methods: All patients underwent bilateral DHL. 3D gait analysis was performed preoperatively, 1 year postoperatively and over 10 years postoperatively. Preoperative temporal parameters, kinematics and GDI were compared with values obtained 1 and 10 year follow-up visits.

Results: The mean age of patients at time of first surgery was 8.3 years (range, 5.4-16.3y), and mean time from first surgery to last 3D gait analysis was 11.8 years (range, 10.0-13.3y). Mean pelvic tilt was not changed significantly after SEMLS including DHL. Mean knee flexion at initial contact decreased from 31.1° preoperatively to 26.0° at 1 year postoperatively (p=0.065), and then decreased significantly to 23.6° at 10 years postoperatively (p=0.038) versus the preoperative value. For the ankle, mean dorsiflexion at initial contact and peak dorsiflexion in the swing phase slightly increased from 1.8° and 9.8° preoperatively to 7.1° and 14.5° at 1 year postoperatively (p=0.059 and 0.469), respectively. However, these kinematic parameters significantly decreased to 1.2° and 7.1°, respectively, 10 years postoperatively. Rotational kinematic parameters such as mean hip rotation and mean foot progression in stance consistently improved over 10 years postoperatively. Mean GDI score significantly improved from 69.4 preoperatively to 77.9 at 1 year postoperatively (p=0.003) and continuously improved to 82.2 at 10 years postoperatively (p=0.017).

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Conclusions: Sagittal knee kinematics, such as, knee flexion at initial contact and knee flexion in terminal swing consistently improved over 10 years postoperatively without increasing mean pelvic tilt. The present study demonstrates that GDI, which represents overall gait pathology, consistently improved after SEMLS, whereas ankle kinematic parameters such as ankle dorsiflexion at initial contact and peak ankle dorsiflexion in swing, slightly improved at 1 year postoperatively and then deteriorated beyond their preoperative values. Single event multilevel surgery including DHL provides a favorable outcome 10 years postoperatively in patients with spastic diplegic cerebral palsy.

A3

Retrospective comparison of age at surgery and rate of remodeling of distal femoral extension osteotomies in children with cerebral palsy

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Background/Objectives: The optimal age to perform a distal femoral extension osteotomy for the treatment of crouch gait in children with cerebral palsy is controversial. In skeletally immature patients, the distal femoral physis will remodel, potentially causing a recurrence of knee flexion deformity. The purpose of this retrospective analysis is to compare the rates of femoral bone remodeling with age at the time of surgery.

Design: Retrospective, non-randomized cohort.

Participants and setting: Following IRB approval, participants with a diagnosis of cerebral palsy who underwent a distal femoral extension osteotomy between January 1999 and April 2013 at our institution were included.

Methods: Nine measures comparing angles between the distal osteotomy and the proximal femur were made on lateral radiographs. A random linear coefficient model was used to ascertain a post-op profile of measures. An interaction of time by age of surgery was introduced into the mixed model to see if age at surgery was related to post-op change in radiographic measures. Results: 69 patients (49 males and 20 females) underwent distal femoral extension osteotomies on 124 legs. Average age at

Table 1

Angle	Average rate of remodeling (deg/mon)	Pearson correlation: age vs remodel rate	P- value
Proximal anterior cortex to distal anterior cortex	0.9184	-0.25618	0.0041
Proximal anterior cortex to distal posterior cortex	0.7907	-0.30786	0.0005
Proximal anterior cortex of the femur and the physis	0.5415	-0.05580	0.5382
Proximal anterior cortex to Blumensaat's line	0.6305	-0.22279	0.0129
Middle of the prox. femur to the middle of the distal femur	0.6769	-0.23486	0.0086
Proximal posterior cortex to distal anterior cortex	0.8607	-0.20326	0.0236
Proximal posterior cortex to distal posterior cortex	0.5288	-0.21167	0.0183
Proximal posterior cortex to physis	0.6455	-0.30208	0.0007
Proximal posterior cortex to Blumensaat's line	0.6176	-0.44468	<.0001

surgery was 13.6 (2.9) years. Slope (rate) was significant for every outcome; this rate was positive, indicating all measures increased over the post-op period. Pearson correlation of estimated slope with age at surgery was negative and significant in 8 of 9 outcomes, indicating the older the Participant had the surgery, the slower the remodeling process of the distal femur after the surgery (Table 1).

Conclusions: An average rate of remodeling was identified for all measures. Older participants with cerebral palsy who underwent a distal femoral extension osteotomy exhibited slower remodeling rates of the distal femur when compared to younger participants in our cohort. To date, this is the largest study looking at remodeling rates of distal femoral extension osteotomies in children with cerebral palsy. The average rate indicates that the correction obtained decreases over time, and this rate differs depending on age at surgery. The data presented here suggests that providers should take the differing rates into account when deciding when and how much correction to obtain in a distal femoral extension osteotomy.

Α4

The frequency of avascular necrosis following reconstructive hip surgery in children with cerebral palsy: a systematic review

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Background/Objectives: Children with Cerebral Palsy (CP) undergoing reconstructive hip surgery are at risk for developing avascular necrosis (AVN). The purpose of this systematic review was to investigate the reported frequency of AVN, and risk factors for developing AVN, following reconstructive hip surgery in children with CP.

Design: Systematic review.

Participants and setting: Studies investigating the outcome of reconstructive hip surgery in patients with cerebral palsy that identified the presence or absence of AVN at a minimum of 12 months post-operative were included. Exclusion criteria were inadequate follow-up, <10 participants in a given study, age at intervention >18 years, and outcome data not clearly separated by surgical technique or primary diagnosis.

Methods: We performed a systematic review of the medical literature using a key word and MeSH heading search strategy. EMBASE and MEDLINE databases were used. Two reviewers selected articles in an independent, stepwise manner. Study quality was assessed using the Methodological Index for Non-Randomized Studies (MINORS) and the Oxford Centre for Evidence-Based Medicine scale.

Results: Three hundred and ninety-nine articles were identified using our search strategy. Twenty-nine studies were included for data extraction after full text review. Many studies were excluded because they did not comment on the presence or absence of AVN. The incidence of AVN ranged from 0% to 46% with a crude rate of 1.66%. A standardized incidence rate, taking into account sample size and years of follow up was 1.72%. Presence of AVN was the primary outcome in two of the 29 included studies. The frequency of AVN in these studies was significantly higher than the majority of the other studies at 37% and 46%. No correlations were found between age at surgery, severity of hip subluxation and the frequency of AVN. There was a trend towards increased rate of AVN with combined varus derotation osteotomy and pelvic osteotomy. There was no significant difference in AVN frequency and length of follow-up. The majority of studies did not comment on methods used for determining diagnosis or severity of AVN. Clinical significance was also not well documented. All studies were of low quality (Oxford Level 4; MINORS scores 4–12/24).

Conclusions: Children with cerebral palsy undergoing reconstructive surgery for hip subluxation or dislocation are at risk of developing AVN postoperatively. Frequency and severity of this complication is poorly documented in the literature. Based on current evidence it is not possible to draw conclusions on risk factors for development of AVN. Incidence of AVN was higher in studies in which AVN was a primary outcome suggesting that the true frequency of AVN may be significantly higher than is currently understood. Prospective multicenter studies will be needed to determine frequency and severity of AVN, the associated risk factors and clinical importance in patients with cerebral palsy undergoing reconstructive hip surgery.

A5

The role of rectus femoris transfer in the development of crouch knee gait in cerebral palsy

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Background/Objectives: The impact of Rectus Femoris Transfer (RFT) at knee flexion during swing phase is very well described, but effects of it in stance phase remains unclear. The aim of this study was to analyze knee flexion during stance phase after rectus femoris transfer, in order to evaluate the influence of RFT in the development of crouch knee gait in cerebral palsy (CP).

Design: Retrospective Cohort Study.

Participants and setting: A total of 3283 patients with spastic diplegic cerebral palsy seen at our gait lab from 1996 to 2013 were identified from our database. Inclusion criteria were: (i) Gross Motor Function Classification System (GMFCS) levels I-III; (ii) kinematic criteria for stiff-knee gait at baseline; (iii) individuals who underwent orthopedic surgery in the lower limbs and had done gait analyses before and after the intervention.

Methods: Patients who fulfilled the inclusion criteria were divided in 2 groups: Group A (185 knees), including patients who underwent orthopedic surgery without a RFT between exams; Group B (123 knees), represented by patients who underwent orthopedic surgery including a RFT. Clinical and kinematic parameters were evaluated at baseline and at follow-up for all groups. The primary outcome was minimum knee flexion in stance (MKFSt) and the secondary outcomes included total knee range of motion (KROM). A statistical analysis was applied with 5% of significance level.

Results: The two groups matched at sex distribution, GMFCS profile, pre-operative GDI, knee flexion in stance phase before surgery and follow-up time (3y). In Group A, the mean MKFSt increased from 13.19° to 16.74° (p=0.003) and in Group B from 10.60° to 14.80° (p=0.001). The post-operative MKFSt was similar among Groups A and B (p=0.534). In Group B, the MKFSt after surgery was higher in patients with GMFCS III (22.51° – p<0.001). A significant increase in MKFSt in the second exam (from 13.01° to 22.51°) was observed only in the GMFCS III patients in the RFT group (p<0.001). An improvement in KROM (from 32.79° to 36.63°) after surgery was noted only in Group B (p<0.001), and this effect was more remarkable for patients GMFCS II.

Conclusions: In this study, RFT was not related to the development of crouch knee gait after a mean follow-up time of 3 years. The increase in MKFSt was similar between Groups A and B. Patients with GMFCS III who underwent multilevel surgery including RFT, exhibited higher increase in MKFSt after treatment than GMFCS levels I and II. The improvement of KROM was observed only in the RFT group, and it was more significant for the GMFCS II patients.

A6

Sensitivity of the CPCHILD questionnaire to change following surgery for scoliosis in children with severe cerebral palsy

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Background/Objectives: Children with severe (non-ambulant) cerebral palsy (CP) often undergo major interventions to preserve or improve their comfort, ease of caregiving, health and quality of life. The Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD) questionnaire was developed to evaluate the effectiveness of these interventions to achieve these goals. It has been shown to be reliable and valid. This study sets out to establish whether the CPHILD is responsive (sensitive to change) to surgery to correct scoliosis in children with severe CP.

Design: Prospective comparative cohort study for outcome measure validation.

Participants and setting: Primary caregivers/parents of 271 children with severe CP (GMFCS III, IV and V) and progressive scoliosis, recruited from 11 centres across the United States and Canada.

Methods: Parents completed the CPCHILD questionnaire at baseline. 202 children (Cases) underwent spinal fusion surgery, and the CPCHILD administered at 6 and 12 months post-operatively. For the 69 (Controls) who did not undergo surgery, the CPCHILD was completed 12 months after baseline. The change in CPCHILD scores from baseline was evaluated in both groups. Responsiveness was analyzed in the Cases by

Paired t-test of the pre-post scores; Standardized Response Means (SRM); and Effect Sizes of the change over 12 months. Results: The groups were comparable at baseline for age; sex distribution; GMFCS level; severity of co-morbidities and CPCHILD scores. Cases had a mean (SD) curve of 85.1° (23.3°) compared with 70.6° (20.1°) for Controls (p<0.05). The CPCHILD Total scores at baseline were 51.2 (14.8) and 54.4 (14.1) in the groups respectively; and also comparable in all six domain (subscales) scores. For Cases the mean (SD) total CPCHILD score was 56.1 (12.7) at 6 months; and 57.0 (13.8) at 12 months after surgery. At 12 months from baseline there were significant improvements (mean differences of +4.9 to +8.7 points) in 5 out of 6 subscales of the CPCHILD for the Cases; whereas in the Controls, total score and subscales remain unchanged or deteriorated (mean differences of -2.8 to +3.3). The SRM ranged from 0.26 to 0.5 for the subscales and 0.45 for the total CPCHILD score; ES ranged from 0.25 to 0.46 for the subscales and 0.37 for the total score.

Conclusions: The CPCHILD is sensitive to change following surgery for scoliosis children with severe CP, with moderately large SRM and ES. The CPCHILD identifies significant improvements in total scores and in the expected subscales, 6 and 12 months after surgery. In a comparable group that did not undergo surgery, scores remained stable or declined at 12 months. The CPCHILD may now be recommended as the primary outcome measure for use in clinical trials or cohort studies of spine interventions for children with severe CP.

A7

Comparison of lumbar epidural bupivacaine with fentanyl or clonidine for postoperative analgesia in children with cerebral palsy after single event multilevel surgery: a double blind randomized clinical trial

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Background/Objectives: Providing effective postoperative analgesia for children with cerebral palsy (CP) undergoing single event multilevel surgery (SEMLS) is challenging because of multiple surgical incisions, muscle spasm and side-effects. Clonidine (BC) or fentanyl (BF) can supplement epidural bupivacaine infusion, but there is no data to guide which regimen is best.

Objectives: To compare epidural BC and BF for differences in postoperative muscle spasm (primary outcome), analgesic efficacy, sedation scores, vomiting or nursing intervention (secondary outcomes) in children with CP undergoing SEMLS. Design: Randomised controlled trial: (double) blinded. Participants and setting: Fifty children in a pediatric tertiary institution.

Methods: After ethics approval and written informed parental consent, consecutive patients were randomised to receive BF or BC epidural solution postoperatively for 72 hours. Continuous variables means were compared using Student's t-test. Non-parametric numerical data were analysed using median±interquartile range (IQR) and the Mann-Whitney U test. Intergroup categorical variables were analysed by c2 test or Fisher's exact test as appropriate from 2×2 contingency tables. Statistical significance was p < 0.05.

Results: Data from 49 patients (BF: n=24, BC: n=25) showed more patients (5/25 vs 0/24) had 2 or more painful muscle spasm epochs in the BF group (p=0.02). There were less spasm epochs (49 vs 8, p=0.01) and fewer patients experiencing spasm (8 vs 3 p=0.01) in the BC group in GMFCS 111/ 1V patients. Fewer BC patients with soft-tissue only surgery had muscle spasm (0/5 vs 3/5, p=0.04). Epidural analgesia was effective in both groups (median pain score 0 [IQR 1]) with fewer epochs of pain requiring intervention in the softtissue only group versus bony surgery (p=0.004). More children in the BF group (19/25 vs 11/24, p=0.03) required two or more epidural boluses. GMFCS III and IV children receiving BC needed fewer epidural boluses (24 vs 46, p=0.03). Children receiving BC experienced less vomiting (mean 0.54/patient vs 2.4/patient; p=0.001), needed less antiemetics (p=0.002) and required less nursing interventions (p=0.01). Two patients in the BF group were withdrawn due to refractory emesis. Patients in the BC group spent less time full alert in the first 24 hours (p=0.001). No patients were judged to be unrousable. Mean systolic and diastolic blood pressure were lower in the BC group. Lower heart rates and more bradycardic (<60bpm) events were recorded in the BC group. There were more epochs of oxygen desaturation <94% in the BF group (in 13 patients, BF 22 vs BC=1, p=0.002).

Conclusions: Clonidine is a superior adjuvant when added to epidural bupivacaine after SEMLS as evidenced by less muscle spasms, vomiting and antiemetic use and reduced need for epidural boluses. Epidural analgesia was safe and effective in both groups. Children in the BC group required fewer nursing interventions related to analgesia.

A8

Lower extremity strength reference curves for ambulatory individuals with cerebral palsy

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Background/Objectives: Individuals with cerebral palsy (CP) are weaker than their age-matched typically developing peers. Strength varies between and within Gross Motor Function Classification System (GMFCS) levels. Strength assessments are frequently used in clinical settings to determine appropriate clinical care and to determine treatment effectiveness. Yet, limited comparison data for lower extremity strength of ambulatory youth with CP, by CP type, GMFCS levels, sex and age are available. Reference curves for strength assessments of these individuals would be useful in prognostication,

intervention planning and assessing progress over time. The study objective was to develop reference curves for strength normalized to body weight (STR-N) for individuals with CP, bilateral involvement, by age, sex and GMFCS level.

Design: Descriptive cohort study.

Participants and setting: Data collected from a convenience sample of 280 individuals (181 male, 99 female) with spastic CP and bilateral involvement, GMFCS levels I-III (I=67, II=136, III=77), ages 8 to 18 years (12y 9mo (2y 8mo) at baseline) from 7 pediatric orthopedic hospitals were included.

Methods: Participants completed an assessment of strength at baseline and 12-months later. Strength of 8 lower extremity muscles bilaterally was assessed with a hand-held dynamometer according to a standardized, published protocol. STR-N is the mean of the maximum strength value for all muscles divided by body weight. Reference centiles were estimated using a generalization of the gamma model from 457 observations accounting for associations between baseline and follow-up measurements. Separate STR-N curves were estimated for boys, girls and GMFCS level. Likelihood ratio tests were used to compare reference curves across GMFCS levels and between sexes within each GMFCS level.

Results: Reference curves were developed for STR-N (Figure 1). Comparisons of curves across GMFCS levels for boys and girls were significantly different (p<0.0001, likelihood ratio test). Significant differences were found between curves for boys and girls within each GMFCS level (p=0.04).

Conclusions: Using a novel approach, STR-N reference curves were generated for individuals with CP (bilateral involvement). Differences between sexes and GMFCS levels are present. These curves can be used to normalize an individual's data to age, sex and GMFCS level of matched peers; to assess, monitor and improve treatment management of ambulatory individuals with CP; and to further aid clinicians in prognostication and intervention planning. An individual's centile can be improved through increases in strength, reduction of body weight or a combination of both. Application of these curves includes assessing associations between strength centiles and functional performance on outcome tools.

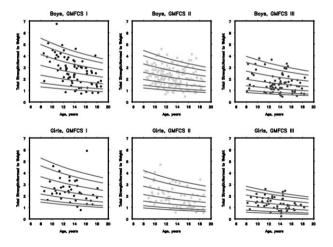


Figure 1: STR-N; lines represent 5th, 10th, 25th, 50th, 75th, 90th and 95th.

A9

Longitudinal change in foot posture in children with cerebral palsy

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Background/Objectives: Foot deformities are common but the evolution of dynamic foot postures is not well documented for children with cerebral palsy (CP). The purpose of this study is to identify unique trends in the development of foot deformity in children with CP from early walking to adolescence using a pedobarograph. Documenting trends in foot posture for children with CP during their growing years will allow clinicians to better understand and treat the underlying mechanisms leading to foot varus and valgus deformities.

Design: Foot pressure was evaluated in children with spastic CP in this IRB approved prospective longitudinal cohort study and compared to age-matched typically developing (TD) children. Participants and setting: Ninety-six children with spastic CP were initially recruited and fifty-one children (16 unilateral, 35 bilateral involvement; 37 GMFCS I/II, 14 III/IV; n=847) met inclusion criteria. We compared this data to age-matched data from 334 feet of TD children.

Methods: Children were initially evaluated at 33(7) months and then evaluated every 6 months until they were 5, and once a year until they were 11 (5 visits minimum for inclusion). We excluded data collected after boney foot surgeries or tendon transfer, but not after soft tissue lengthening surgery. For each visit, the average of 3 dynamic foot pressure measurements was analyzed using the F-Scan measurement system (Boston, MA). Outcome measures included the coronal plane pressure index (CPPI) and heel impulse. CPPI is a ratio of the medial and lateral pressure impulses in the midfoot and forefoot regions. ANOVA with Tukey post-hoc tests were used to compare the groups. Data were grouped and analyzed graphically using a thin plate spline analysis package for R statistical software.

Results: Variability in foot pressure data was higher in the children with CP than it was in the TD group. From age 3 to 11 years, CPPI was higher (valgus) in children GMFCS III/ IV compared to TD (p<0.05 for all except age 7), and this was

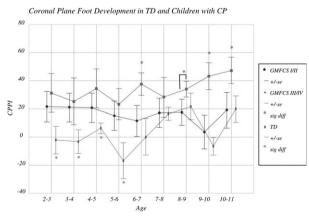


Figure 1: Strength centile versus GMFM centile.

consistent over time (Figure 1). In the GMFCS I/II group, CPPI was higher than TD from age 3 to 5 (p<0.05), but then decreased to the normal range. Heel impulse was reduced in both GMFCS I/II and III/IV groups compared to TD regardless of age (p<0.05), and the III/IV group had less heel contact than the I/II group (p<0.05).

Conclusions: The development of dynamic foot posture in walking is highly variable in children with CP especially at early ages. Young children with CP tend to have a valgus foot distribution relative to TD. Valgus tends to persist in children with GMFCS levels III and IV and to normalize in children with GMFCS levels I and II. Due to variability in the natural history of foot posture in children with CP, conservative management of coronal plane foot deformity is suggested, especially in young children and those ambulating without an assistive device. Further research is needed to identify the specific factors that influence these abnormal trends in foot posture development in children with CP.

A10

Direct quantification of passive muscle stiffness in children with cerebral palsy using shear wave elastography

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Background/Objectives: In children with cerebral palsy (CP), passive muscle stiffness appears to be abnormal due to structural differences including fewer sarcomeres in series and increased collagen. Passive muscle stiffness is important as it contributes to the force generation of the muscle, development of contracture, and is a target of antispasticity treatment. Previous studies have tried to address passive muscle stiffness, but due to measurement limitations, have not been able to isolate the muscle (without influence of joint capsular or tendon stiffness) and have been limited to older ambulatory children. Therefore, we propose to use a new ultrasound technique, shear wave elastography (SWE), to noninvasively and directly quantify passive muscle stiffness. Our aim is to demonstrate feasibility of use of this tool in children with CP and compare passive muscle stiffness of the lateral gastrocnemius muscle in children with CP to typically developing (TD) children.

Design: Cross-sectional study.

Participants and setting: 20 TD children (2y 0mo-11y 6mo, 12 girls, 8 boys) and 9 children with CP (2y 1mo-9y 3mo, 4 girls, 5 boys, GMFCS level I-IV) were recruited from the community and a quaternary care medical center.

Methods: With the child prone, SWE measurements of bilateral lateral gastrocnemius muscles were performed at area of greatest calf circumference. Each foot was passively positioned to 20° plantarflexion, 10° plantarflexion, 0° plantarflexion. This was repeated for a total of 3 trials. The Aixplorer® (Super Sonic Imagine, Aix en Provence, France) with a linear

Table 1: Demographics and physical exam measurements of the typically developing children (TD) and children with cerebral palsy (CP).

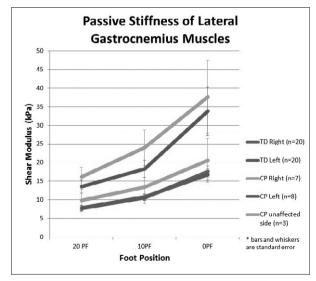
		TD (20)	CP (9)	p-value⁴
Age * (months)		81.1 (38.0)	66.1 (31.6)	0.3105
BMI* (kg/m²)		18.9 (4.5)	16.3 (2.0)	0.1221
Calf Circumference*	Right	27.2 (5.6)	21.0 (4.1) ^b	0.0180
(cm)	Left	27.2 (5.6)	21.9 (3.0) ^c	0.0256
Maximum Ankle	Right	13.0 (7.18)	2.9 (11.3) ^c	0.0107
Dorsiflexion* (degrees)	Left	13.2 (6.3)	1.0 (10.1) ^d	0.0006
GMFCS Level (n)	1	na	2	
	П	na	4	
	Ш	na	2	
	IV	na	1	
	V	na	0	
CP Type (n)	Hemiplegic	na	3	
	Diplegic	na	4	
	Triplegic	na	1	
	Quadriplegic	na	1	

wo-tailed t-test with significance at p<0.05, b n = 6, c n=7, d n=8

Table 2: Mean (SE) of the passive stiffness of lateral gastrocnemius muscle of the unaffected side in children with hemiplegic cerebral palsy (CP unaffected side), typically developing children (TD), and affected side in children with cerebral palsy (CP).

Shear Modulus (kPa)	CP unaffected side (3)*		TD (20)	CP (9)	p-value ^a (TD vs CP)
20 PF	9.9 (3.0)	Right	7.7 (0.6)	16.2 (2.5)b	0.0001
		Left	7.8 (0.7)	13.5 (1.7) ^c	0.0012
10 PF	13.5 (4.5)	Right	10.6 (0.8)	24.0 (4.8) ^b	0.0002
		Left	10.7 (0.9)	18.4 (2.3)°	0.0007
0 PF	20.5 (5.9))	Right	17.6 (1.5)	37.6 (9.7) ^b	0.0029
		Left	16.8 (1.5)	33.8 (6.7) ^c	0.0013

 $^{^8}$ two-tailed t-test with significance at p<0.05, n n=7, c n=8 a Due to small numbers in CP unaffected slide, left and right were grouped together. No further statistical calculations were performed on this group.



TD = typically developing children, CP = affected limb in children with cerebral palsy, CP unaffected = unaffected limb in children with hemiplegic cerebral palsy. PF = degre plantarflexion, DF = degrees of dorsiflexion, kPa = kilopascals

US probe, SL15-4MHz, with 'musculoskeletal' preset mode was used to obtain shear modulus measurements. Surface EMG, UControlTM (Thought Technology Ltd., Quebec,

Canada), at lowest setting to detect muscle activation was used to monitor muscle relaxation.

Results: Demographics are listed in Table 1. At all positions, there was a significant difference in shear modulus between the children with CP and TD children (Graph and Table 2). Lateral gastrocnemius passive muscle stiffness increased with increasing stretch with the children with CP having greater stiffness at all positions. In children with hemiplegic CP, the unaffected limb had slightly greater stiffness than the TD children, but less stiffness than the affected limb in CP.

Conclusions: There is significantly greater passive muscle stiffness in children with CP as compared to TD children. In children with hemiplegic CP, there is suggestion of a slight increase in passive stiffness in the unaffected side, which may be due to influence of the affected side or underlying subtle neurologic differences of this side. This tool appears feasible for quantifying passive muscle stiffness in children with CP, including very young children and children who are nonambulatory. This study is the first step toward larger studies quantifying passive muscle stiffness in children with CP longitudinally and evaluating effect of antispasticity treatment, rehabilitation, and orthopedic interventions on this muscle property.

Free Papers B

B1

Quantitative analysis of lower extremity adipose tissue distribution in children with myelomeningocele

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Background/Objectives: Children with myelomeningocele (MM) tend to be more overweight and obese and often have altered body composition compared to youth without MM. The health risks associated with excess adiposity depend largely on location. Children with MM not only have greater abdominal obesity, but they also have more fat accumulating in their lower extremities. However, it is not known if this is subcutaneous (SC) or intramuscular (IM) fat, the latter of which has been associated with insulin resistance¹. Therefore, the purpose of this study was to evaluate the distribution of fat in the lower leg of children with MM.

Design: Prospective, Cross-sectional.

Participants and setting: 21 participants with MM (12 males; 10y 8mo, SD 1y 8mo) and 36 healthy controls (21 males; 10y 3mo, SD 2y 0mo) from a tertiary children's hospital.

Methods: Tissue volumes were computed along the entire length of both tibias from computed tomography (CT) scans. Tissue volumes were quantified using a semi-automated, threshold-based method with fixed attenuation ranges of [-190, -30], [-29, 150] and [151, 1000] Hounsfield units for adipose, muscle and bone, respectively. Fascial boundaries were determined using automated interpolation of manually delineated fascial boundaries in periodic slices. Adipose tissue was classified as intra- or intermuscular (IM) if it was enclosed

within the fascial boundary or subcutaneous (SC) if not. Clinical data gathered included height, weight, body mass index (BMI), Tanner stage of sexual maturity, and International Myelodysplasia Study Group (IMSG) classification. Tissue volumes were compared among controls, sacral level MM, and lumbar level MM using multivariate linear regression.

Results: After adjusting for height and weight, mean IM fat volume increased significantly across control, sacral and lumbar groups (152, 425, 1215cm³, p<0.05), while muscle volume decreased (3037, 2077, 1061cm³, p<0.05) and SC fat volume did not significantly change (2120, 2114, v 2124cm³, p>0.05). In the same model, total adipose volume was higher in the lumbar compared to control groups (3340 vs 2271cm³, p<0.01) but not significantly different in the sacral compared to control groups (2539 vs 2271cm³, p=0.06) or the sacral compared to lumbar groups (3340 vs 2539cm³, p=0.51).

Conclusions: The results of this study suggest that increases in lower leg adiposity in children with MM are primarily due to accumulation of IM fat, while SC fat volumes remain more or less unchanged. Equally striking is the observed decrease in muscle volume associated with MM. Since IM fat is more strongly associated with negative health outcomes than SC fat¹, children with MM may have an increased risk of adverse health effects. Therefore it may be beneficial to consider the anatomical distribution of adipose tissue when treating children with MM.

Reference: 1. Goodpaster, B et al. 'Thigh adipose tissue distribution is associated with insulin resistance in obesity and in type 2 diabetes mellitus'. AJCN 71.4 (2000): 885–892.

B2

Myelomeningocele: mortality risk factor analysis from a national in-patient database

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Background/Objectives: Children with myelomeningocele, the most common type of spina bifida, are frequently hospitalized for various medical problems throughout childhood. A previous 2000 national database study evaluated surgical repair of myelomeningocele. The goal of this study was to examine the impact of patient and hospital characteristics on mortality rate during inpatient stays in children with myelomeningocele using the 2009 version of this inpatient database.

Design: Records review.

Participants and setting: Patient data was collected from the Healthcare Cost and Utilization Project Kids' Inpatient Database.

Methods: Data was collected retrospectively from the Health-care Cost and Utilization Project Kids' Inpatient Database, a resource designed to analyze pediatric hospital usage. The 2009 database includes data from 4121 hospitals in 44 states with a total of 7 370 203 pediatric discharges for inpatient cases. 5769 complete records were identified using ICD-9 codes 741.00 to 741.03 or 741.90 to 741.93 in the 25 diagnosis

fields. This population included 55 patient deaths. The effect of various predictor variables on all causes of mortality during hospitalization was evaluated using logistic regression. Examined variables included: elective procedure, surgery during the admission, patient age, the number of concurrent diagnoses, the number of procedures performed, patient race, sex, income, state, length of stay, payer, teaching-hospital status, and whether the hospital was a children's hospital.

Results: The 55 deaths in this study constituted 0.9% of the cases. Patients <1 year old had a mortality rate of 3.1% (p<0.001, OR 0.83-0.92). However, neonates had the highest mortality risk of 5.1% (p<0.05, OR 1.03-3.84). Patient history also had an effect on outcome. Those with a higher number of overall diagnoses and who had multiple procedures during their stay were more likely to die during hospitalization (p<0.001, OR 1.22-1.48, and p<0.001, OR 1.49-1.70, respectfully). The following variables had no effect on mortality: sex, race, geography, payer, median household income of zip code, number of chronic conditions, hospital location, and teaching status of the hospital. The mortality in the subgroup of patients <1 year of age decreased by referral to a children's hospital as compared to either a children's unit in a general hospital or a general hospital, independent of other factors (0.9%, 3%, 4.4%, respectfully, *p*=0.013).

Conclusions: The results of this study suggest that the mortality rate is highest for neonates and children <1 year of age. As might be expected, mortality was higher in patients with more concurrent diagnoses and more procedures performed. Younger children (<1y) with myelomeningocele requiring hospitalization as an inpatient have a lower mortality risk if referred to a children's hospital. The specific cause of death in these children is not identified in this national database.

B3 Summary data from the national spina bifida patient registry 2009-2012

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Background/Objectives: Surveillance and key practice parameters are important to better understand the natural progression of chronic conditions that present at birth and continue through the lifespan. The Centers for Disease Control and Prevention (CDC) developed a nationwide registry of persons with spina bifida. This abstract provides an overview of the data collected by the participating sites.

Design: Informed consent was obtained from patients/parents of those with spina bifida at these clinics to collect historical and current data based on 20 questions related to demographics and condition management.

Participants and setting: Ten clinics began enrollment of patients into the registry in 2009, and 9 additional clinics began in 2011.

Methods: Data collected is entered into a web-based electronic medical record maintained in a HIPPA compliant IRB approved status at the CDC.

Results: This analysis includes data from 3737 individuals during the initial time of the study 3/09 - 12/12. 9.6% are <2 years; 15.1% 2 to <5; 22.5% 5 to <10; 11.1% 10 to <13;</pre> 20.7% 13 to <18; 11.2% 18 to <22; and 9.9% 22 or older, at last contact. Race and ethnicity: 64.6% Non-Hispanic white. 7.3% Non-Hispanic black, 21.4% Hispanic or Latino, and 6.7% other. 51.5% have public and 47.8% have private insurance, and 0.7% are uninsured. Spina bifida type: 81.5% have myelomeningocele and 18.5% have a variant of non-myelomeningocele. Functional levels of lesions: 15.5% thoracic, 9.0% high lumbar, 27.2% mid-lumbar, 19.3% low lumbar, and 28.9% Sacral. Current student's education: 18 to 21 years: 56.7% primary/secondary, 5.9% technical school, 26.4% some college, 1% college, and 10% other; 22 yrs and older: 15.8% employed full time, 17.7% employed part time, 31.5% not employed/permanently disabled, 21.7% were able to work but not currently employed, and 8.2% were not employed students. 65.1% had a shunt placed for hydrocephalus. 88.1% had bladder and 80.5% had bowel impairment. Bladder management methods for those 5 years and over: 72.9% clean intermittent catheterization, 14.8% spontaneous void, 3.4% vesicostomy, 1.5% indwelling catheter, 0.7% urostomy, 0.1% Credé, and 8.4% none. Urinary continence defined by dry during the day was reported by 43.7% and bowel continence defined by no involuntary stool leakage was reported by 50.6% in those 5 years and older. Urologic surgeries included bladder augmentation (12.4%), continent catheterizable channel (12.6%), and vesicostomy (5.1%). For aged 2 years and older, 54.6% were described as community, 8.6% as household, 7.9% as therapeutic ambulators and 28.9% as nonambulatory. Females represent 51.8%.

Conclusions: Preliminary information from the largest spina bifida database in the world, including 3737 individuals treated in 19 multidisciplinary clinics in the United States, is useful in describing demographics and management of this condition. Continued utilization and expansion of this registry should help to better define best clinical practices, improve care and identify disparities in treatment outcomes.

B4

Development of gross motor function in children with cerebral palsy: an investigation of 'motor arowth curves'

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Background/Objectives: In Norway children with Cerebral Palsy (CP) are systematically followed up with both specialized and general health care. The CP- follow-up program (CPOP) was implemented in South-Eastern Norway in 2006 and nationally in 2010. The main aims of CPOP are to generate knowledge about gross motor function of CP, provide a predictable follow-up program for the families and prevent functional limitations in children with CP. The children are assessed according to a standardised protocol once or twice a

year or every second year depending on age and GMFCS level. The protocol includes: Diagnosis according to SCPE, GMFCS and GMFM-66. The objectives of the present study was to describe patterns of gross motor development by level of severity of CP according to GMFCS and GMFM-66 total score in the Norwegian population of children with CP and compare the Norwegian motor growth curves with The Ontario Motor Growth Study (OMG) (Hanna et al 2009) and a corresponding study from Nederland (Smits et al 2013).

Design: A longitudinal population-based study.

Participants and setting: The children in CPOP are assessed with GMFM-66 and number of assessments varied from one to nine. The GMFCS classification and GMFM-66 assessments are performed by physical therapists at 21 Habilitation units.

Methods: The registrations are saved in a database at Oslo University Hospital. The data are analysed with descriptive statistics in SPSS 18.

Results: Totally 592 children, 246 girls, from 1 to 11 years, are assessed with 1488 GMFM-66 observations. CP subtypes; 45% bilateral CP, 42% unilateral CP, 8% dyskinetic CP, 4% ataxic CP, 1% not classified. GMFCS classification: Level I 45%, Level II 17%, Level III 11%, level IV 12%, level V 15%. The Norwegian study had no exclusion criteria for interventions, such as spasticity reducing treatments and orthopaedic surgery. When comparing the Norwegian motor growth curves with the Ontario Motor Growth study (OMG) and the resent Dutch study there seems to bee a relatively stable development up to 9 years for the five growth curves with mean score on GMFM as described in Table 1.

Table 1

GMFCS level	1- 5 years	5- 9 years	9 -11 years
I	67,2	84,2	90,3
П	52,0	70,7	73,7
III	45,1	52,6	53,6
IV	32,3	42,6	40,8
V	20,3	22,9	22,4

Conclusions: The present study confirms the Canadian and Dutch studies on motor growth curves. Further research is needed to follow the Norwegian children up to adult age.

B5 Assessment of the association between strength and function in youth with CP using newly developed reference curves

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Background/Objectives: Youth with cerebral palsy (CP) are weaker than typically developing peers; how strength impacts function is unclear. Gross motor function of children with CP is often evaluated using measures normed on children without

motor impairments. Reference curves for strength and outcome tools based on youth with CP stratified by age, sex and severity would improve the ability to determine how a child is functioning. This study uses newly developed reference curves for strength normalized to body weight (STR-N) and outcome tools to assess the association between strength and function in youth with CP.

Design: Descriptive cohort study.

Participants and setting: Data from a convenience sample of 280 individuals (181 male, 99 female) with bilateral spastic CP, GMFCS levels I–III (I=67, II=136, III=77), ages 8 to 18 years (12y 9mo (2y 8mo) at baseline) from 7 pediatric orthopedic hospitals were included.

Methods: Participants completed assessment of GMFCS, strength, walking speed, GMFM-66, Pediatric Outcomes Data Collection Instrument (PODCI), modified Timed-up-and-Go (mTUG) at baseline and 12-months later. STR-N is the mean strength of 8 lower extremity muscles assessed bilaterally with a hand-held dynamometer divided by body weight. Reference centiles were estimated using a generalization of the gamma model from 457 observations accounting for associations between baseline and follow-up measurements. Separate STR-N curves were estimated for sex and GMFCS. Curves for outcome tools were separated only by GMFCS level as differences between sexes were not significant (walking speed [p=0.72], GMFM-66 [p=0.37] and PODCI [p=0.79]). Using the reference curves, each individual's strength and outcome measures were converted to centiles. Correlation coefficients were used to assess associations between strength and outcome tool centiles. STR-N data were grouped into quartiles and plotted against outcome tool centile (Figure 1). Kruskal-Wallis test was used to compare distribution of outcome centiles across strength quartiles.

Results: Median values for outcome score centile significantly increased with increasing strength quartiles for GMFM-66 (p<0.001) and walking speed (p=0.013). Figure 1 shows GMFM-66 data; graphs for all other outcome tools are similar. Significant correlations were found between all strength and outcome centiles (Table 1).

Conclusions: Using newly developed reference curves for STR-N and functional assessments a fair positive association

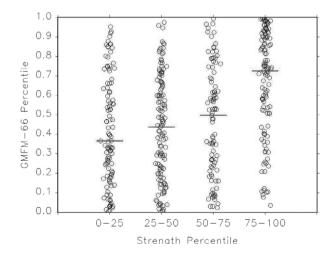


Figure 1: Strength centile vs GMFM centile.

Table 1: Correlations between strength centile and outcome score centile.

Tool	R	95% CI	p-value
GMFM-66	0.35	0.27, 0.43	< 0.001
Walking Speed	0.15	0.06, 0.24	0.002
PODCI Global Function	0.10	0.00, 0.19	0.045
mTUG	-0.17	-0.28, -0.07	0.002

between STR-N centile and function was seen. Within a GMFCS level stronger individuals tend to have higher outcome tool centiles. These findings suggest if an individual's STR-N centile improves, improvements in outcome tool centile is possible.

B6

Building a model to address the role of parenting in the lives of children with neurodevelopmental disorders (NDD): does overprotectiveness matter?

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Background/Objectives: A major assumption in practice is that overprotective parenting is problematic as it limits childrens' ability to engage with their environment, thereby compromising personal growth. Surprisingly, little is known about how robust this assumption is. This exploratory study unpacks this phenomenon by examining relationships between complexity of the NDD, parental overprotectiveness, and child mastery motivation.

Design: A cross-sectional clinical study Parenting Matters! was conducted and exploratory analysis used to address the nature of the relationships between these variables.

Participants and setting: A convenience sample of 236 parents (mothers: n=197; age range 4–12v; girls: 37.9%) with an NDD child were recruited from tertiary care pediatric sites in Alberta, Ontario and Quebec, Canada. 21% of children had a diagnosis of ASD, 31% CP, 20% Epilepsy, and 28% Other. Mean complexity of disability was 13(5.3) (on a scale ranging from 0 to 26).

Methods: Parents completed self-report measures that evaluated demographics, complexity of their child's disability, parenting and their child's mastery. Complexity of child's disability was measured with About My Child (AMC; Rosenbaum et al., 2008), a 26-item instrument that evaluated function (e.g., mobility, communication, cognitive ability); overprotection with 4 subscales of the 25-item Parent Protection Scale (PPS; Thomasgard et al., 1995): supervision, separation problem, dependence and control; mastery with the Dimensions of Mastery Questionnaire (DMQ; Morgan et al., 2009), which has 7 subscales (object-oriented persistence, social persistence with adults, social persistence with children, gross motor persistence, mastery pleasure, negative reactions, and general competence).

Results: The first analytic stage revealed associations between complexity of the child's diagnosis and parental supervision (r=0.24; p=0.01) and control (r=0.33; p<0.001). The only relationships that were significant between parenting and mastery were between the supervision dimension of parenting and a child's social persistence with children (r=-0.20; p=0.01) and mastery pleasure (r=-0.18; p=0.02). When mastery pleasure was treated as the dependent variable, neither complexity of disability nor parenting, were significant (p>0.05). The same was true when social persistence was the dependent variable. However, when parenting (supervision or control) were treated as dependent variables, complexity was a significant predictor (*B*=0.19; *p*<0.0001; *B*=0.09; *p*=0.05).

Conclusions: Complexity of the disability may inform how protective and engaged parents are in their parenting relationship. However, neither complexity of disability nor parenting have much to do with how a parent experiences their child's sense of mastery motivation or social persistence. These findings suggest that we should be cautious about recommending that parents be less 'overprotective' as our assumptions about the difference that it will or will not make to this aspect of child well-being is not empirically validated.

B7

Caregiver burden in cerebral palsy is related to disease severity and life stages

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Background/Objectives: Caring for a child with a disability can be stressful. Through the Learn From Every Patient (LFEP) program at Nationwide Children's Hospital, extensive data on each child are collected during each visit to the Interdisciplinary CP Program. Several standardized assessments are completed annually, including the Caregiver Burden Scale (CBS).

Design: Retrospective Cohort Study.

Participants and setting: Interdisciplinary CP Clinic Patients.

Methods: We retrospectively examined the data from the CBS, a validated survey that asks about stress, financial burden, and relationship dynamics. The survey was administered in clinic by the Social Worker. The Gross Motor Classification Scale (GMFCS) was assigned by the physical therapist and the Communication Functional Classification Scale (CFCS) by the Speech therapist. Pierson correlation coefficients and ANOVA were used for analysis.

Results: Data were available on 306 children from 2012 to 2013. Their mean age was 6.84 years ([4.73]; range 0-27). 32% of the patients were classified as GMFCS level 1, 15% level 2, 10% level 3, 23% level 4 and 20% level 5. 23% of the patients were classified at level 1 on the CFCS; 15% level 2; 13% level 3; 21% level 4 and 28% level 5. Although communication and gross motor abilities individually only accounted for 6% (p=0.00) of the variance seen in total survey scores,

there was a significant difference between stress in families of children with less gross motor impairment (levels 1 and 2) and those with more severity (levels 4 and 5) (t=-3.53, df=273, p<0.00) Age was not significantly correlated to the total survey scores. When grouping the ages into preschool (0–5y), elementary school (6–10y), middle school (11–14y), and high school (15–18y) a peak in stress was reported for the school age group. A second smaller peak of stress is seen with the transition out of high school. Highest levels of stress were worrying about the child's pain, the financial impact due to lost income, and limits family activities. Lower stress was reported for financial impact of medications, doctor's visits, and causing arguments or tension in the family.

Conclusions: Caregivers of severely affected children with CP reported greater burden than caregivers of children with milder disabilities. The greatest sources of stress were the child's pain and family functioning related to employment and activities. It is encouraging that caregivers reported minimal burden related to costs of doctor appointments and medications. We believe that this means that we (as a program, hospital, and society) are doing a good job of connecting families with financial support resources. An increase in reported stress was noted at transition ages. Sources of this stress can include difficulties planning appropriate educational services, understanding special education law, and the Individualized Education Planning process. Increased education and advocacy through the Comprehensive CP Program should be further assessed to determine how to address this need.

B8

A longitudinal examination of anxiety and depression among youth with spinal cord injury: the importance of caregiver and community factors

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Background/Objectives: Little is known about how anxiety and depression change over time among youth with spinal cord injury (SCI), and in particular how contextual factors impact mental health as children age. The current study examined changes in anxiety and depression over time among youth with SCI and how anxiety and depression relate to child (age, sex, injury level and duration), family (caregiver education), and community (type) factors.

Design: Longitudinal survey study.

Participants and setting: At the time of first interview, youth were ages 7 to 18, had sustained a SCI at least 1 year prior to enrollment, and were receiving care from Shriners Hospitals for Children in Chicago, Philadelphia, or Northern California.

Methods: Youth were approached annually to complete standardized measures of anxiety (all youth) and depression (youth

ages 7–17). Primary caregivers (typically parents) completed a project-specific demographics form. Data from 597 annual interviews from 321 youth with SCI were analyzed using linear mixed models. 321 youth contributed one interview, 184 two interviews, 75 three interviews, and 17 four interviews. Data were analyzed using Proc Mixed in SAS.

Results: At the time of first interview, the 321 youth were an average age of 13.67 years (SD=3.48) and they had been injured an average of 5.06 years (SD=4.33); 57% were male, 65% had paraplegia, and 52% lived in small town/rural areas. 63% of caregivers had at least some college experience. Age, injury duration, caregiver education, and community type contributed significantly to anxiety. In particular, youth who were younger, youth with more recent injuries, youth whose caregivers had less education, and youth living in city/urban/suburban areas had greater anxiety. Age, caregiver education, and community type contributed significantly to depression. In particular, younger youth (ages 7-8) and older youth (age 17) and youth whose caregivers had less education experienced greater depression. Further, an interaction between age and community type revealed that small town/rural youth experienced greater depression at a young age than city/urban/suburban youth.

Conclusions: Age, injury duration, caregiver education, and community type are important predictors of the mental health of youth with SCI. These findings support Urie Bronfenbrenner's ecological systems theory of child development, in that various family and community factors are impacting youth outcomes. Results suggest that intervention efforts should strive to treat the whole child and his/her family in context.

B9

Evaluation of the disparity of harm caused to children with medical complexity while hospitalized

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Background/Objectives: Pediatric patients have been shown to be susceptible to medical errors in the hospital setting. Children with medical complexity (CMC) comprise one of the most rapidly growing subsets of pediatric patients. These patients may be at increased risk for errors in their care due to the complex nature of their medical conditions. The study objectives were to determine if CMC sustain increased harm while hospitalized compared to those patients without medical complexity, and to determine the incident types and severity of adverse events occurring in hospitalized CMC.

Design: We conducted a retrospective study examining the possible disparity of harm to patients with medical complexity at Nationwide Children's Hospital. A CMC was defined as any patient who had >2 inpatient discharges in 1 year and >2 diagnoses per discharge over the study period. For each adverse event reported to the institution's Event Reporting

System (ERS), a rating of one through nine on a Clinical Severity Scale (CSC) was assigned.

Participants and setting: Nationwide Children's Hospital is a 450 bed tertiary hospital in Columbus, Ohio, with a catchment area of 37 counties. Approximately 650 000 children live in these counties, while patients come from all 50 states and internationally. All patients aged 0 to 18 years discharged from the hospital from July 2009 to September 2012 were included in the study.

Methods: The CSC is an adaptation of the 2001 National Coordinating Council for Medication Error Reporting and Prevention (MERP) Scale. Harm which reached the patient as a result of the adverse event was defined as any event rated as a level four or higher in the ERS. By comparing the incident types and severity of adverse events between CMC and patients without medical complexity, it can be determined if CMC are more likely to suffer harm while hospitalized. Descriptive statistics were used to describe the population and a 2 Sample Poisson Rate was used to test the difference in rate of harm in CMC and children without medical complexity.

Results: Children with medical complexity made up 27% of inpatient discharges and accounted for 64% of hospital days. Their Average length of stay was more than twice that of children without medical complexity and total hospital days were five times greater. 27% of CMC had a patient safety event compared to 8% of children without medical

	Nationwide Children's Hospital Clinical Severity Scale
1	Circumstances or events that have the capacity to cause error or harm
2	An event occurred but did not reach the patient
3	An event occurred that reached the patient but was not followed by patient harm
4	An event occurred that was followed by increased patient monitoring and/or minimal patient harm or minor injury
5	An event occurred that was followed by treatment or intervention and/or temporary patient harm
6	An event occurred that was followed by initial or prolonged hospitalization and temporary patient harm
7	An event occurred that was followed by permanent harm
8	An event occurred that was followed by near-death event
9	An event that was followed by patient death

Source of Scale: Nationwide Children's Hosptial adaptation of the 2001 National Coordinating Council for Medication Error Reporting and Prevention (MERP) Scale

Note: Only PREVENTABLE events are reported to the Harm Index Categories

Figure 1: Nationwide Childrens Hospital Clinical Severity Scale.

	Children with Medical Complexity	Children without Medical Complexity	Total
Patients	10,201 (26.9%)	27,703 (73.1%)	37,904
Visits	21,900 (40.9%)	31,683 (59.1%)	53,583
Hospital Days	211,904 (64%)	119,242 (36%)	331,146
Avg. Length of Stay (ALOS)	10	4	6
Avg Visits Per Patient	2	1	1
Hospital Days Per Patient	21	4	9

Figure 2: Hospitalization characteristics of CMC and children without medical complexity.

by Clinical Severity & Patient Type 25% **■ Children w/o Medical Complexity** 20% Percent of Inpatients 15% 10% 5%

Percent of Inpatients with Safety Events

Figure 3: Percent of CMC and children without medical complexity with a safety event by clinical severity.

0%

Misses

Events (Sev 1-3) (Sev 4-9)

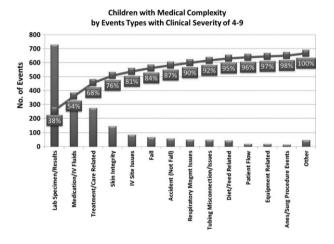


Figure 4: Pareto of safety events with harm for children with medical complexity.

complexity (OR=3.3). CMC were more likely to have a patient safety event that caused harm as compared to children without medical complexity (p<0.001). On average, CMC who had a safety event were likely to have two safety events. 68% of harm events for CMC were related to Lab Specimen/ Results, Medication/IV Fluids, and Treatment/ Care.

Conclusions: CMC are 3.3 times more likely to have an adverse event reported than those patients who are not complex; are more likely to have multiple adverse event reports; and are more likely to have been harmed by these adverse events. Error reduction strategies should be targeted towards CMC.

B10

Parent, therapist and researcher perspectives regarding important participation elements for children who use power mobility using a Delphi survey

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Background/Objectives: Power mobility interventions help overcome participation restrictions in everyday life for children with significant mobility limitations. Existing health measurement scales differ in how they evaluate participation. However, limited research exists on what elements of participation are relevant and important to measure for children who use power mobility.

Design: An on-line Delphi survey to establish consensus among an expert panel on elements of participation deemed important to measure for children who use power mobility. Consensus was set *a priori* at 80% agreement.

Participants and setting: Purposive sampling was used to recruit an international expert panel including parents whose child, aged 18 months to 12 years, used power mobility at least weekly for >6 months, therapists with >5 years experience in pediatric power mobility with >5 children seen in last year, and researchers with >3 years experience in power mobility or participation with children having physical disabilities.

Methods: The first round identified elements of participation important to measure in two age cohorts: preschool children aged 18 months to 5 years, and school-aged children 6 to 12 years. The second round confirmed elements of importance and the third round prioritized these elements.

Results: A total of 74 participants included 14 parents from Canada and the United States, 40 clinicians from 10 countries and 20 researchers from 8 countries. Consensus was reached on 8/48 elements for preschoolers and 18/48 elements for school-aged children. For preschoolers: Parents, therapists and researchers rated measuring participation in a combination of settings as most important, however parents rated child's enjoyment higher and barriers and facilitators lower than the other two groups. For school-aged children: Therapists and researchers agreed on the top four priorities: measuring a combination of settings, a combination of parent and child reports of participation, child's engagement and barriers and facilitators to participation. Parents' highest priority was child's enjoyment, followed by barriers and facilitators, a combination of settings, and combination of parent and child reports.

Conclusions: Experts achieved consensus on 26 elements of participation perceived as important to measure for preschool and school-aged children who use power mobility, although priorities varied among parents, therapists and researchers. These elements should guide decision-making when choosing

a suitable measure for this population. Differences between preschool and school-aged children and different priorities among parents, therapists and researchers merit consideration when evaluating participation outcomes. These findings reinforce the importance of a family-centered approach when evaluating outcomes of children's power mobility use.

Free Papers C

C1

Validity of the OMNI- Rate of Perceived Exertion scale for youth with cerebral palsy

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Background/Objectives: Therapy interventions often focus on increasing physical activity (PA) frequency and intensity to promote health and mobility in youth with cerebral palsy (CP). PA intensity can be measured by oxygen consumption (VO2), heart rate (HR), or accelerometry; however, these methods may not be practical in community settings due to cost or measurement burden. Perceived exertion rating scales are low cost, feasible options for measuring PA intensity. The OMNI Rate of Perceived Exertion Scale (OMNI-RPE) has not been validated for youth with CP. The aim of this study was to evaluate the criterion validity of the OMNI-RPE with HR and VO2.

Design: Cross-sectional, Instrument Validation Study.

Participants and setting: A sample of 57 ambulatory youth with

CP, GMFCS Levels I-III, ages 6 to 18 years were recruited from two pediatric hospitals. Children were excluded if they had recent orthopedic surgery or botulinum neurotoxin injections. Methods: Youth with CP(GMFCS Level I; 50%), mean age 12.6 (SD=3.3) completed 9 PA trials: supine resting, sitting/ writing, wiping a counter, folding laundry, playing X-Box Kinect games (River Run, Space-Pops) and walking at 3 selfselected speeds (slow, brisk, and fast). Participants wore a portable indirect calorimeter and HR monitor while performing the trials, each lasting 2.5 to 6 minutes. Participants received standardized instructions on the use of the OMNI-RPE walk/ run scale and they reported their RPE at the completion of each trial. Within-subject correlations between OMNI-RPE, HR and VO2 over the 9 trials were calculated. For the whole sample and for each of the GMFCS level groups, within subject correlations were averaged using Fisher-z transformations. GMFCS level group differences in the correlation coefficients were evaluated. A Friedman 2 way ANOVA was used to determine if OMNI-RPE could distinguish intensity across trials.

Results: Forty-six participants had complete VO2 data, HR data, RPE for all 9 trials. Median OMNI-RPE values with 25th to 75th centile ranges were supine resting=0 (0–0), sitting/writing=2(0–2), wiping a counter=2(0–3.5), folding laundry=2(0.25–4.75), River Run=4(1–5), Space Pops=4(2–6), slow

walk=3(1.25–6), brisk walk=4.5(2–9), and fast walk=8(4–10). For the total sample GMFCS Levels I-III, VO2 (r=0.73; CI 95 0.56–0.84) and HR (r=0.76; CI 95 0.58–0.86) were moderately correlated to OMNI-RPE scores. Correlations were lower for youth classified at GMFCS Level I for VO2 to OMNI-RPE (r=0.63; CI95 0.30–0.82) and HR to OMNI-VO2(r=0.67; CI 95 0.31–0.86); however, no significant differences were found when comparing results between GMFCS levels. OMNI RPE was able to distinguish intensity across trials (F8, 360=66.61, p<0.001). Post hoc pairwise comparisons indicated that OMNI-RPE increased with PA intensity in a dose-response manner.

Conclusions: The OMNI- RPE may be a valid option to monitor relative PA intensity in youth with CP classified at GMFCS levels I-III.

C2

Is it feasible to use the Gross Motor Function Measure-66 Basal and Ceiling score in children with acquired brain injury?

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Background/Objectives: The Gross Motor Function Measure version-88 (GMFM-88) and version-66 (GMFM-66) have good psychometric properties and are the criterion standard measure of gross motor function for children with cerebral palsy, but are also recommended for use in children with acquired brain injury (ABI) (McCauley et al. 2012). Both measures are time intensive to complete (Russell et al. 2002); therefore the Basal and Ceiling (GMFM-66-BC) and the Item Set (GMFM-66-IS) were developed. However, the suitability and psychometric properties of the GMFM-66-BC and the GMFM-66-IS in children with ABI are not known. Therefore this study aimed to compare the GMFM-66-BC to the GMFM-66 in children with ABI, whilst identifying any factors that may affect the feasibility of using the GMFM-66-BC in children with ABI.

Design: Longitudinal retrospective validation study of the GMFM-66-BC in children with ABI.

Participants and setting: An existing data set of 277 GMFM-88 assessments from 76 participants (mean age 11y, SD 5y, median 11y 11mo, range 10mo–17y 6mo) were obtained between October 2009 and May 2013 from all children with ABI who attended a specialist in-patient rehabilitation centre (Surrey, UK). Strict inclusion and exclusion criteria were applied to each GMFM-88 assessment to ensure suitability for use in GMFM-66 and GMFM-66-BC formats. This resulted in 119 GMFM-88 assessments, from 49 children with ABI (mean age 11y 8mo, SD 4y 6mo; 25 males) across all Gross Motor Function Classification System levels.

Methods: GMFM-88 data sets were converted into GMFM-66 and GMFM-66-BC formats to enable the level of agreement and association between the GMFM-66-BC and GMFM-66 to be compared at single and between multiple time points. The GMFM-66-BC expected level of item difficulty for this population of children with ABI, was also evaluated against the GMFM-66-BC order of item difficulty. The GMFM-66

expected level of item difficulty was determined by calculation of the expected scores (Russell et al. 2002).

Results: At single and between multiple time points Bland Altman plots showed minimal systematic difference in agreement (mean<0.05, p>0.44) and the overall association between GMFM-66 and GMFM-66-BC was good ($r\geq0.81$). However, the order of item difficulty identified within GMFM-66-BC, as calculated for children with CP, differed from the expected level of item difficulty when calculated for this population of children with ABI.

Conclusions: The GMFM-66-BC is potentially a feasible outcome measure for children with ABI due to the accuracy of its estimates. However, the suitability of the order of item difficulty identified in the GMFM-66-BC and GMFM-66 mean that further research with a larger data sample is required before recommending either measure for use in children with ABI.

C3

The development and validation of the Fatigue Impact and Severity Self-Assessment scale for use with youth and young adults with cerebral palsy

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Background/Objectives: There are many scales available to measure fatigue; however, none have been validated for use in a population of individuals with cerebral palsy (CP). A well-constructed, valid and reliable tool to assess fatigue in CP would assist therapists and clients in collaborative goal setting and intervention planning throughout the lifespan, enhance self-awareness and self-management of CP.

Design: This was an instrument development and validation study.

Participants and setting: This study was conducted in two phases. Participants for the first phase consisted of health care professionals across multiple disciplines that regularly treat individuals with CP. Participants from the second phase of the study included individuals with CP aged 14 to 31 recruited from Ontario children's treatment centres, the general community and from online postings in virtual communities related to CP.

Methods: Potential items for the Fatigue Impact and Severity Self-Assessment (FISSA) were generated from a systematic review of fatigue measures and a phenomenological study of interviews with individuals with CP. Focus groups were then conducted with health care professionals who regularly treat individuals with CP, to reduce the items contained in the scale and to add items to reflect any missing domains. A large sample of participants (n=130) completed the FISSA to assess reliability and discriminative validity. Internal consistency was assessed using Cronbach's alpha and test-retest reliability was assessed over 2 to 4 weeks with a small subset of the participants (n=31) using an ICC. Tests of known-groups validity using the Gross Motor Function Classification System (GMFCS) and pain were used to establish the construct validity of the FISSA. An explor-

atory factor analysis provided information about the factor structure of the scale.

Results: Cronbach's alpha for the entire questionnaire was 0.95. The FISSA demonstrated an ICC(3.1)=0.74 (95% CI 0.53–0.87). Individuals who self-classified as GMFCS level I experienced less fatigue than individuals in any other GMFCS level (II–V) (*p*<0.001). Individuals with higher pain (both impact and severity) reported higher fatigue scores (*p*<0.001). The FISSA contains 31 items related to two factors (Impact of Fatigue and Management and Activity Modification) that together explain 48.7% of the variance in fatigue scores.

Conclusions: The FISSA was created to examine the severity, impact and management of fatigue for youth and young adults with CP. The FISSA is reliable and was able to discriminate between groups expected to experience more fatigue including those with a more severe motor disability according to the GMFCS and individuals with a high degree of pain. The FISSA allows for individualized identification of the activities of daily living that may be compromised by fatigue, which may enhance collaborative goal setting and intervention planning by clinicians and their clients.

C4

Using ActiGraph® accelerometers in children and adolescents with congenital hemiplegia: days of monitoring needed and test-retest reliability

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Background/Objectives: To establish the variability in measurement of physical activity, and test-retest reliability of a triaxial accelerometer in independently ambulant children with congenital hemiplegia.

Design: Reliability measurement study.

Participants and setting: Children and adolescents with congenital hemiplegia (n=95; 48 males; 49 right hemiplegia; mean age 11y 3mo [2y 4mo], range 8–17y) classified at Gross Motor Functional Classification System (GMFCS) I=41 and II=54 within the community environment. To establish test-retest reliability, 30 children were recruited via consecutive series sampling (16 males; 13 right hemiplegia; mean age 11y 11mo, range 8–17y; GMFCS I=16, II=14) and participated in a research laboratory.

Methods: Variability: Participants wore an ActiGraph®GT3X+tri-axial accelerometer for 4 days. ActiGraph® activity counts were converted into activity intensity using uni-axial derived cut points to classify the time spent in moderate-to-vigorous physical activity (MVPA). Between-day intra-class reliability coefficients (R) and Spearman-Brown prophecy formula ([ICC_desired/1-ICC_desired][1-ICC_estimated/ICC_estimated]) were calculated to determine daily variability in MVPA. Test-retest reliability: Participants completed standardized tasks over two consecutive days wearing an ActiGraph®GT3X+ accelerometer and heart rate monitor while video recording. Testing protocol comprised 5 minutes of seated rest (REST), walking at light (LW), moderate (MW) and vigorous (VW) pace, and

rapid stepping on/off a step (STEP). Two minutes of synchronized data was extracted for each task. Reliability was calculated between days using intra-class correlation coefficients (ICC; two-factor mixed agreement model). Minimum detectable difference was calculated (MDD=[SD $\sqrt{1}$ -ICC] $\times 1.96 \times \sqrt{2}$).

Results: Of a potential 380 days, 357 days (94%) were collected. Three to 4 days of monitoring produced acceptable variability estimates of MVPA (R=0.70 [0.57–0.79] to 0.73 [0.0.61–0.81]). Spearman-Brown prophecy analysis estimated 3 days were needed to achieve a reliability coefficient of 0.7, and 11 days to achieve 0.9. Activity intensity increased with standardized task, as reflected by axis-1 activity counts and heart rate (figure 1). Test-retest reliability was strong for walking tasks (ICC=0.70–0.80), but moderate for STEP tasks (ICC=0.66). Agreement between activity intensity was generally strong (80–98%), but during MW reduced to 17%. MDD was between 1214 and 2139 counts/minute.

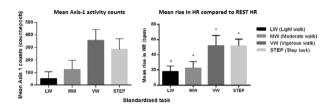


Figure 1: Mean Axis-1 activity counts and rise in heart rate by standardised task intensity.

Conclusions: At least 3 days of monitoring is necessary to produce reliable estimates of MVPA in independently ambulant children with congenital hemiplegia. The ActiGraph® tri-axial accelerometer has moderate to strong test-retest reliability under controlled walking and stepping tasks in this population. Accelerometers can reliably measure physical activity capacity under controlled conditions, as well as performance in a community environment.

C5

Use of goal attainment scaling in post-acute care setting

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Background/Objectives: Goal attainment scaling (GAS) has been used widely in rehabilitative settings. It is a patient centered means of setting goals and prioritizing them according to a patient's wishes rather than a preconceived idea of what is most important medically. We felt that the use of GAS would be beneficial in 2 ways: (i) it would focus care on goals that were important to the patient and family and (ii) for patients at the extremes of functional scales such as WeeFIM—gains may be difficult to see because of floor and ceiling effects. In these cases, GAS goals may help illustrate gains that were important to the family but not picked up by standardized instruments.

Design: All patients (if capable) and their parents were asked what their goals were. This was illustrated by saying, what do you hope we accomplish in this hospitalization. These were recorded on the care plan that was begun at admission. At the first interdisciplinary meeting, therapists would discuss what they felt was possible in terms of the goals and adjustment was made as described by Lynne Turner-Stokes. Every 2 weeks progress toward the patient's goals was recorded.

Participants and setting: Participants were 1046 inpatients admitted to a post-acute care hospital between 1/1/2012 and 12/31/2013.

Methods: Each unit of the hospital had a coordinating pediatrician and it was this physician who asked the patient and family about their goals.

Results: Percentage of goals achieved was 59.6%. In terms of the team's ability to predict success, they averaged 50.9 which is ideal. This will be explained further in presentation. Importantly, even when the Wee FIM failed to show significant improvement, the GAS enabled families to set achievable goals that were appropriate for their child.

Conclusions: Gas was readily accepted by the treatment team. The pediatricians felt that it brought them closer to the family on the first day. They also felt that they gained valuable information about why a particular child was admitted. Even for patients with purely medically complex problems, asking about their goals was felt valuable in terms of ascertaining parents' insight into their child's problem.

C₆

Bimanual fine motor function classification in children with cerebral palsy: aspects of content and construct validity

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Background/Objectives: During the last decade two classification systems describing functional hand use in children with cerebral palsy (CP) have been developed; the Bimanual Fine Motor Function (BFMF) and the Manual Ability Classification System (MACS). Both the BFMF and the MACS aim to classify hand function in children with CP on a five level scale. However, the purpose of the two classification systems differs. The purpose of the MACS is to describe manual performance and it is regarded a valid and reliable classification of how children with CP handle objects in daily life. The purpose of the BFMF is to describe fine motor function by classifying ability to grasp, hold and manipulate objects. Thus these two

classification systems may complement each other. The MACS is used extensively both in clinical practice and in research, while the BFMF is used in several CP registers established by the Surveillance of Cerebral Palsy in Europe (SCPE). Inter-rater reliability of the BFMF was recently found to be high, but the classification system has not yet been validated. The aim of this study was to explore aspects of content and construct validity of the BFMF.

Design: Validating cohort study.

Participants and setting: Population-based data from 539 children born 1999 to 2003 (304 boys; 4–12y) were extracted from the Norwegian Cerebral Palsy Register and from the Cerebral Palsy Register of western Sweden.

Methods: Construct validity of the BFMF was explored by investigating the correlation between children's BFMF and MACS levels. In addition aspects of content validity of the BFMF was explored by making a judgment about the relevance and comprehensiveness of the items through literature review, and by using the International Classification of Functioning, Disability and Health-Children and Youth (ICF-CY) as a framework to describe how the different constructs of the BFMF and the MACS are interrelated.

Results: Construct validity of the BFMF was supported by high correlation with the MACS (Spearman's ρ =0.89, CI: 0.86–0.91, p<0.001). Judgment about the relevance and comprehensiveness of the BFMF items suggest that the ability to hold, grasp and manipulate seems to describe increasingly advanced fine motor abilities in children with CP. When the content of the BFMF and the MACS were linked to the ICF-CY, they were both found to capture fine hand use in the component activity and participation. Whereas the MACS was found to reflect manual performance, it could not be determined from the content of the BFMF whether it is a classification of fine motor capacity or performance.

Conclusions: Our results suggest that the BFMF is a valid classification of differing levels of fine motor function in children with CP. If provided a more specific instruction and further validation, the BFMF have the potential to classify fine motor capacity. Thus, the use of the BFMF in addition to the MACS may provide useful information regarding the potential difference between fine motor capacity and actual use of the hands in daily life.

C7

Psychometric properties of the revised Assisting Hand Assessment version 5.0

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Background/Objectives: The Assisting Hand Assessment (AHA) is a commonly used test which measures and describes how effectively children with unilateral disability in arm and hand use their affected hand in bimanual tasks. The AHA has been widely used for children with unilateral cerebral palsy (UCP). The first version of the AHA containing 22 items was published in 2003 and even though it has shown to have sound psychometric properties and to be sensitive to change, experience

from almost 10 years of use called for a revision. The objective of this study was to scrutinize the AHA test items for possible improvements, revise them and to evaluate the psychometric properties of the revised version of the AHA, version 5.0.

Design: Instrument Development & Validation Study.

Participants and setting: One-hundred and sixty-four assessments of children with UCP aged 18 months to 12 years were included in the analysis. The children were a clinical convenience sample from out-patient rehabilitation services.

Methods: All items were scrutinized for the use of all four steps of the rating scale and four items were alternatively worded to enable improved distribution across the categories. One new item was formulated. Data was analysed using the Rasch measurement model analysis. Internal scale validity was investigated by evaluating, rating scale functioning, item and person goodness-of-fit and unidimensionality of the scale. Furthermore, targeting and scale reliability was evaluated.

Results: After removal of misfitting items a 19 item scale showed acceptable goodness-of-fit. Thus, 95% item fit and 96% person fit to the model was achieved. Unidimensionality was confirmed by a principal component analysis. For revised and remaining items the rating scale was well functioning, indicating that scale steps were used in the intended manner. The item difficulty was well suited to the ability level of the sample. Scale reliability analyses showed a high person reliability coefficient of 0.98, indicating high separation ability of the scale. Compared to the current AHA 4.4 version, the 5.0 is somewhat shorter but has improved internal scale validity and person reliability and is thereby promising to be more sensitive to change. Information making comparison between outcomes of current version 4.4 and the new version 5.0 possible will be made available to AHA-users.

Conclusions: The revised version of the AHA shows evidence of excellent psychometric properties, being more sensitive for change as well as being somewhat shorter to score.

C8

Test-retest reliability of the assessment of motor and process skills in 8- to 16-year-old children with unilateral cerebral palsy

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Background/Objectives: The Assessment of Motor and Process Skills (AMPS) is a standardized, observational evaluation of the motor and processing skills involved in the performance activities of daily living (ADL) tasks. This study investigated the test-retest reliability of the AMPS in children with unilateral CP aged 8 to 16 years.

Design: Reliability measurement study.

Participants and setting: Thirty children (mean age=11y 7mo [2y 4mo]; males=18; Manual Ability Classification System (MACS) I=15, II=15; Gross Motor Function Classification System (GMFCS) I=9, II=21) enrolled in a large randomized controlled trial were recruited via consecutive series sampling.

Methods: The AMPS was undertaken in a naturalistic domestic area that was free from distractions in a clinical setting. The retest interval was set at 2 days. On day 1, the occupational therapist conducted an AMPS interview and participants selected two tasks from a list of appropriate task options (e.g. Task 1: Making a jam sandwich and Task 2: Preparing breakfast cereal and a cup of juice). The participants then carried out the task according to standardized AMPS procedures. On day 2, participants performed the same two tasks under the same conditions. The children's tasks were video recorded on both days and later scored and entered into the AMPS software by an AMPS calibrated occupational therapist. Data were analysed using intraclass correlation coefficient (ICC) two-way random, single measures model (2.1) for the AMPS motor and the AMPS process scales (SPSS v22). The minimal detectable change (MDC) was calculated using the formula: $(SD\sqrt{1-ICC})\times 1.96\times \sqrt{2}$.

Results: Test-retest reliability was excellent on both the AMPS motor scale (ICC=0.93; 95% CI=0.86-0.97) and AMPS process scale (ICC=0.86; 95% CI=0.65-0.94). Intra-rater reliability (n=10) was excellent for AMPS motor scale (ICC=0.96; 95% CI=0.81-0.99) and AMPS process scale (ICC=0.98; 95% CI=0.75-0.99). The MCD was 0.23 for the AMPS motor scale and 0.30 for the AMPS process scale.

Conclusions: This study found excellent test-retest reliability for both the AMPS motor and process scales in children with unilateral CP. Activities of daily living motor skills appear marginally more stable than processing skills. Individuals are less likely to vary in their motor abilities on a day to day basis but may fluctuate in the way they order steps and modify their actions. The AMPS can be used with 8 to 16 year old children with unilateral CP with changes in scores >0.23 on the AMPS motor scale and 0.30 on the AMPS process scale reflecting real changes in ADL motor and processing abilities.

C9

Measuring the discriminant validity of the PEDI-CAT in children with cerebral palsy

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Background/Objectives: Patients with Cerebral Palsy (CP) demonstrate a broad range of physical and cognitive impairments, making the reliability of any one functional assessment tool challenging. The Pediatric Evaluation of Disability Inventory-Computer Adaptive Test (PEDI-CAT) utilizes a computer adaptive platform with an increased item bank of 276 items divided into domains of (i) Daily Activities; (ii) Mobility; (iii) Social/Cognitive, and (iv) Responsibility. The objective of this study was to measure the discriminant validity of the PEDI-CAT according to motor function in a diverse population of children with CP.

Design: IRB approved, Prospective Cross-Sectional Convenience Sample (Level III).

Participants and setting: Sixty-nine English-speaking children/ parents with CP were recruited from our tertiary level, multidiscipline children's clinic between August 2013 and January 2014. Children were excluded if their diagnosis was uncertain or if they had undergone recent surgery (<6mo) or botulinum neurotoxin A administration (<3mo).

Methods: Age, sex, race, Gross Motor Function Classification Level (GMFCS), Functional Mobility Score (FMS), and Manual Ability Classification Score (MACS) were recorded for all participant families. Items from the 4 PEDI-CAT domains (Daily Activities, Mobility, Social/Cognitive and Responsibility) were administered to parents via an iPAD and GMFCS, FMS and MACS level were scored by treating physicians at the time of PEDI-CAT application. Differences in mean PEDI-CAT scores were tested using an analysis of variance across the five GMFCS and MACS levels. Spearman correlations were used to assess the association between FMS and PEDI-CAT scores. Our primary hypothesis was that the PEDI-CAT would be able to differentiate levels of mobility according to the GMFCS, FMS and MACS. Our secondary hypothesis was that an inverse relationship exists between each PEDI-CAT domain score and GMFCS level.

Results: There were 13 GMFCS I (19%), 17 GMFCS II (25%), 8 GMFCS III (12%), 13 GMFCS IV (19%), and 18 GMFCS V (26%) children who participated in this study. Mean age was 11.7 years (SD +3.7). All four domains of the PEDI-CAT were able to differentiate levels of mobility according to GMFCS (p<0.001) with Spearman correlations ranging from -0.90 (Mobility) to -0.62 (Responsibility/ Social/Cognitive) (p<0.001). The PEDI-CAT domains were also able to differentiate between MACS levels (p<0.001) with higher correlations ranging from -0.93 (Daily Activities) to −0.75 (Responsibility) (p<0.001). The PEDI-CAT Mobility scores demonstrated high correlation with FMS 5m (r=0.89, p<0.001) and FMS 50m (r=0.86, p<0.001), and moderate correlation with FMS 500m (r=0.74, p<0.001). An inverse relationship existed between PEDI-CAT domain scores and GMFCS level (r=-0.62 to -0.90, p<0.001).

Conclusions: This study demonstrates that the PEDI-CAT is able to discriminate between function and mobility levels across a population of children with CP and an inverse relationship exists between PEDI-CAT scores and GMFCS levels.

C10

A measure of youth self-reported experiences of activity settings (SEAS)

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Background/Objectives: Little is known about how youth experience their community/home leisure activity settings. There is an identified need for self-report measures of experiences of participation, rather than those completed by proxies. The 22-item SEAS is a youth-rated measure of experiences of personal growth, psychological engagement, social belonging, meaningful interactions, and choice and control. The SEAS is designed for youth with and without disabilities who have a

grade 3 level of language comprehension or more, including youth with severe disabilities.

Objective: To describe the measure's development and psychometric properties.

Design: Instrument Development and Validation Study.

Participants and setting: Five content experts took part in the consultant review phase, with backgrounds in psychology, AAC, speech-language pathology, occupational therapy, and child/adolescent psychiatry. Pretesting, piloting, and psychometric testing involved youth with and without disabilities. In the psychometric phase, 45 youth ages 14 to 23 (10 with severe disabilities) completed the SEAS in 160 leisure activity settings. The SEAS was developed at a children's rehabilitation centre in Toronto Canada.

Methods: Development involved: (i) item generation based on the literature; (ii) consultations with experts, including youth with disabilities, parents, and scholars; (iii) pretesting and piloting; and (iv) psychometric testing. Scales were determined using principal components analyses. Test-retest reliability was determined for 8 youth with severe disabilities who completed the SEAS twice (an interval of 2–4wks) for the same type of activity setting.

Results: The factor structure accounted for 62% of the variance. The KMO Measure of Sampling Adequacy was 0.79 and Bartlett's Test of Sphericity was significant (p<0.0001). Cronbach's alphas for the scales (Personal Growth, Psychological Engagement, Social Belonging, Meaningful Interactions, and Choice and Control) ranged from 0.71 to 0.88. Test-retest reliability (mean scale ICC=0.68) was moderate, as expected due to changes in activity settings over time. The SEAS was able to differentiate group from solitary activity settings, with group settings rated higher in Psychological Engagement than solitary ones (M=0.46, SD=1.41 and M=-0.33, SD=1.81, respectively, t(65)=2.61, p<0.001; t=0.32).

Conclusions: Results indicate the SEAS has a sound factor structure and preliminary evidence of excellent internal consistency and adequate test-retest reliability. The SEAS can be used to gain greater understanding of the situation-specific experiences of youth participating in various types of leisure activity settings. The SEAS will be useful for the design of social ecological programs for youth with disabilities, and for research on associations between youths' experiences and qualities of their environments.

Free Papers D

D1

Prevention of baclofen withdrawal syndrome: pharmacokinetics and tolerability of oral and intravenous baclofen in healthy adult volunteers

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Background/Objectives: Patients treated with oral or intrathecal baclofen (ITB) may experience a withdrawal syndrome when

Table 1: Mean ± SD of baclofen pharmacokinetic parameters following oral (10mg) and intravenous (5mg) administration in healthy volunteers.

Pharmacokinetic Parameter ^a	5 mg IV Mean ± SD	10 mg Oral Mean ± SD
C _{max} (ng/mL)	313 ± 75	176 ± 15
T _{max} (hr) ^b	-	1.0 [0.5-2.0]
AUC _{last} (ng.h/mL)	587±100	825 ± 132
AUC _{0-∞} (ng.h/mL) ^c	719 ± 169	1033 ± 202
Bioavailability (%)		74± 15
T _{1/2} (hr)	5.4 ± 1.6	5.0 ± 1.4

- a: Mean values are presented as arithmetic means
- b: Median (Min, Max) reported for T_{max} c: Two-tailed p value < 0.05 (Paired t-test performed on dose-normalized AUC)

therapy is acutely interrupted. The management of baclofen withdrawal is inadequate with slow response and frequent adverse effects secondary to therapy. An intravenous (IV) baclofen formulation might prevent or minimize withdrawal symptoms but is not available. Study aims were to characterize pharmacokinetics (PK) and safety of baclofen given orally and IV in healthy participants.

Design: Twelve adult healthy participants were enrolled in a randomized, open-label, crossover study after IRB approval. Participants and setting: Participants involved in this study were 18 to 65 year old healthy volunteers with BMI between 19

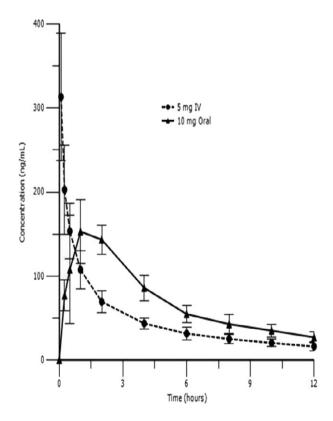


Figure 1: Mean±SD plasma concentration- time profiles of baclofen after intravenous and oral administration in nine participants (0-12h).

and 30kg/m^2 . The volunteers were medication free for 48 hours before, during, and 24 hours after the administration of the study drug.

Methods: Participants received single doses of baclofen: 3 or 5mg given IV (Lioresal Intrathecal®, 2mg/mL) and 5 or 10mg taken orally with a 48-hours washout. Blood samples for baclofen analysis were collected pre-dose and at regular intervals up to 24 hours post-dose. Plasma baclofen concentration-time data were analyzed using non-compartmental PK methods. Descriptive statistics were used to summarize PK parameters and a paired t-test was used to test for significant difference in IV versus oral area under the curve (AUC). Safety was assessed by ECG, blood pressure and pulse monitoring; assessment of CNS toxicity; injection site irritation; side effects and tolerability; as well as physical exams at 12 and 24 hours after the drug administration. In addition, sedation using Sanford sleepiness scale, ataxia, and nystagmus were assessed.

Results: Mean concentration-time data of nine participants who received a 5mg IV dose and a 10mg oral dose on separate days are shown in Figure 1. The mean absolute bioavailability of oral baclofen was 74% (95% CI: 61%, 86%) (Table 1). There was a significant difference in dose-adjusted AUCs (p=0.0024) between oral and IV arms. AUC variability was similar (CV: 18-24%) in both arms. Adverse effects were mild in severity and not related to either dose or route of administration. Most common adverse effects were somnolence, mild ataxia and nystagmus, all of which were resolved within 6 hours after drug administration.

Conclusions: Three and 5mg doses of IV baclofen were well tolerated and 74% oral bioavailability indicates that smaller doses of IV baclofen will be needed to attain total drug exposure comparable to oral dosing. The PK data from this study will guide design of future clinical trials that are required for commercial development of IV baclofen.

D2 Intrathecal baclofen therapy versus orthopedic surgery in adolescents with cerebral palsy in **GMFCS** level III

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Background/Objectives: Multilevel orthopedic surgery and intrathecal baclofen therapy (ITB) are considered to treat children with cerebral palsy (CP). This retrospective study aims to evaluate the effects of these different therapies in children with spastic diplegia in GMFCS level III, usually very impaired due to spasticity and musculoskeletal deformities.

Design: Cohort study (retrospective).

Participants and setting: We considered a population of patients with a diagnosis of spastic diplegia, <18 years old, classified by GMFCS level III. A total number of sixty patients with a mean age of 12.2 years (SD=3.5) were evaluated. Thirty children received lower extremity orthopedic surgery and thirty received ITB.

Methods: At baseline and 12 months after surgery evaluated by muscle tone (Ashworth scale), motor function (Gross Motor Function Measure [GMFM-88]), gait analysis, functional musculoskeletal health (Pediatric Outcomes Data Collection Instrumental [PODCI]) and a subjective questionnaire.

Results: The analysis revealed that GMFM total score, dimensions D and E significantly increased in both groups at the follow up (p<0.05). Spasticity (p<0.001) and normalized spatio-temporal (p<0.05) scores significantly improved only in ITB patients. All PODCI scores significantly improved in ITB group while in orthopedic surgery group only upper extremity and basic mobility changed significantly (p<0.05). The subjective questionnaire showed an over all satisfaction with both the therapies.

Conclusions: ITB therapy and orthopedic surgery both improve motor function and mobility, have different advantages and provide different results (spasticity reduction and deformities correction). Before administering one or the other treatment the specific awaited objectives for each child need to be defined. For patients in Level III affected by spasticity and lower extremity deformities, the therapeutic algorithm should consider both tone reduction and orthopedic surgery. A possible approach consists in administering ITB therapy and later orthopedic surgery. According to our experience benefits provided by the ITB can limit the need of later surgery.

D3

Is there difference in the proprioception sense of children with right and left hemiplegic cerebral palsy?

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Background/Objectives: Children with hemiplegic Cerebral Palsy (CP) had not only motor dysfunctions but also had impairment in sensorial functions. It is determined that proprioceptive sense deficits had both sides of the body in hemiplegic CP and their proprioceptive sense error greater than their healthy peers. Researches have showed that left hemiplegic children had more severe involvement in motor function, gait pattern, attention and cognitive skills than right hemiplegic children. This study is aimed to investigate difference in proprioception between left and right children with hemiplegic CP.

Design: Crossover study design.

Participants and setting: Twenty children with hemiplegic CP were investigated; of these children 10 had right hemiplegic CP (RHG: right hemiplegic group; age: 8.48 [1.08]y; sex: 4 girls, 6 boys) and 10 had left hemiplegic CP (LHG: left hemiplegic group; age: 9.39 [1.58]y; sex: 5 girls, 5 boys). Proprioception of the children were measured with Prosport 1000 PMS a reliable and valid proprioception measurement device. The inclusion criteria were (i) aged between 5 and 16 years (ii) no range of motion limitations was found. The exclusion criteria were any orthopedic surgery or Botulinum neurotoxin injection in the past 6 months.

Methods: Spasticity of knee flexors, ankle plantar flexors and shoulder flexors were assessed with Modified Tardieu Scale. Joint position sense of lower and upper limb were measured with Prosport 1000 PMS. Upper limbs tests were applied on supine position with shoulder flexion and lower limbs tests on sitting position with knee extension. The selected method of testing was passive reproduction of a target angle. Hemiplegic side shoulder and knee joints of all the participants were evaluated with Johnstone pressure splint at start positions (45 internal rotation, for shoulder; 90 knee flexion for knee) with an angular rotational movement at a constant speed of 0.5°/s. Target angel was determined as 45°. The angular displacements from the target angles at the end of the reproduction tests were recorded as position sense deficit scores. SPSS version of 21 for Macintosh was used for statistical analysis and p-values of 0.05 and less were considered evidence of statistically significant findings. Mann Whitney-U test was used to compare joint position sense of LGH and RGH.

Results: The median (minimum-maximum) value of lower limb deficits was 2.00° (0.66° – 3.16°) in RGH and 3.83° (1.13° – 6.60°) in LGH. The median (minimum-maximum) value of upper limb deficits was 2.55° (1° – 17.66°) in RGH and 8° (4.33° – 9.66°) in LGH. There were statistically significant differences in joint sense position between RGH and LGH (respectively; U:14 000, p=0.006; U: 21 000 p=0.027).

Conclusions: This study showed that proprioception deficits in left hemiplegic children were greater than right hemiplegic children. Therefore, clinic physiotherapists should much consider proprioceptive training in left hemiplegic children with CP.

D4

Baseline gross motor classification versus pediatric quality of life in patients recruited into a randomized, double-blind placebo-controlled study of abobotulinumtoxina (Dysport®) in the treatment of dynamic equinus deformity in children with cerebral palsy

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Background/Objectives: The Gross Motor Function Classification System (GMFCS) has been in use since 1997. To date, little has been published in scientific literature about the quality of life in ambulatory patients with cerebral palsy (CP). The objective of this analysis was to explore the relationship between GMFCS level I to III with the subscales and total score of PedsQL in this population.

Design: A large randomized, double-blind, placebo-controlled, multicentre study (N=241) aiming to assess the long term efficacy and safety of AbobotulinumtoxinA (Dysport®) has recently completed enrollment.

Participants and setting: Children with dynamic equinus foot deformity due to CP receiving multiple treatments of Dysport®

Methods: Ambulatory patients aged 2 to 17, with dynamic equinus deformity, GMFCS (E & R) I–III, Modified Ashworth Scale (MAS) \geq 2, Tardieu Scale $X\geq$ 10°, $Y\geq$ 2, no fixed contractures, previous surgery or phenol/alcohol injections are included in the study. Analysis of baseline GMFCS levels and PedsQL (general and CP module) were performed.

Results: A total of 241 patients, aged 2 to 17 (median 5) years were randomized in the study. There were 133 (55%), 82 (34%) and 26 (11%) of randomized patients with GMFCS I, II and III, respectively. Fifty-nine per cent of patients were male. Fifty percent of patients were hemiplegic and 44% were diplegic. The median baseline MAS scores in ankle plantar-flexors were grade 2 in all 3 GMFCS levels (I–III).

A trend of lower scores in GMFCS III was observed in most subscales of PedsQL in CP module. It is most notably in the CP daily activities where the mean (SD) scores were 66 (28), 48 (27) and 33 (22) for GMFCS level I, II and III, respectively. Both Physical Health and Psychosocial health sub-scale in the general module shows lower scores in patients with GMFCS III patients. In the CP module, School Activities, Movement and Balance, Eating activities and Speech communication subscales were lower in patients with GMFCS III compared to patients with GMFCS I and II. No real difference was observed for the Pain and Hurt subscale, with scores of 85 (19), 83 (19) and 80 (15) for GMFCS I, II and III, respectively.

Conclusions: Ambulatory CP patients with dynamic equinus deformity and GMFCS level III have lower scores in most sub-scales in Peds QL and CP modules, except in the subscale of pain and hurt. The analysis also shows that the majority of ambulatory patients with CP are relatively free of pain.

D5

The immunological response to botulinum neurotoxin-A in toxin-naïve children with cerebral palsy: a randomized clinical trial

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Background/Objectives: To investigate incidence and clinical significance of immunological response to botulinum neurotoxin-A (BoNT/A) in toxin-naïve children with spastic cerebral palsy (CP).

Design: Randomized clinical trial.

Participants and setting: Thirty-eight toxin-naïve, ambulant children (GMFCS I – III, mean age 3y 4mo, SD 1y 1mo) with spastic CP were recruited through two tertiary pediatric hospitals and randomised to receive either 12-monthly (n=20) or 4-monthly (n=18) BoNT/A injections for spastic equinus, over a 26-month period.

Methods: BOTOX® 6U/kg was injected into gastrocsoleus with a total dose of 18U/kg available for other muscles. Blood samples were drawn prior to each BoNT/A injection, de-identified and investigated for binding antibody (AB) by radioimmune assay and blocking AB by mouse protection assay (MPA) specific to the study and separate from the MPA methodology used for the BOTOX® label. Response relationship to the participant's human leukocyte antigen (HLA) genotype was determined and compared to a larger BoNT/A-treated cervical dystonia cohort to determine the frequency of HLA allele type in MPA-positive participants. Clinical resistance to BoNT/A was explored using ankle kinematics prior to and 4 weeks following final injection.

Results: Two participants in the 4-monthly group and none in the 12-monthly group demonstrated non-dose dependent MPA-positive results. Both children were clinically unresponsive to BoNT/A on kinematic testing. Antibody binding was increased but not significant (p=0.18 Student's t-test), in the 4-monthly (1680cpm, SD 2497) compared to the 12-monthly (830cpm, SD 692) group. HLA typing revealed two frequent DQ-allele types, which were also found in one MPA-positive participant. Combination of these with the cervical dystonia data showed higher frequency (p=0.038 Fishers exact test) of DQA1*0102 in the blocking AB-positive group (0.41) than the AB-negative group (0.24) before correction and almost double the frequency (0.20) expected for the predominant Caucasian cohort.

Conclusions: This is the first investigation of immune response to BoNT/A in toxin-naïve, children with CP. Protective-AB were present in two of the 18 children receiving BoNT/A on a 4-monthly basis for 2 years and none of the 18 children receiving BoNT/A on a 12-monthly basis for 2 years. These two MPA-positive samples elicited higher AB bindings to BoNT/A and were clinically unresponsive. Linkage of particular DQA1*0102 with protective AB-positive group was marginally significant with further work in progress and clinical relevance yet to be determined. Small subject numbers with two non-responding MPA-positive participants reflect preliminary data with further research indicated. Immunological resistance to BoNT/A in children with CP may be relevant to regular BoNT/A therapy during childhood.

D6

An electromyographic protocol that distinguishes spasticity from dystonia

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Background/Objectives: The purpose of this study is to demonstrate that electromyographic (EMG) activation patterns of leg muscles differ predictably among patients with predominantly spasticity, patients with predominantly dystonia, and typically developing control participants during rest, active movement, and passively induced movement.

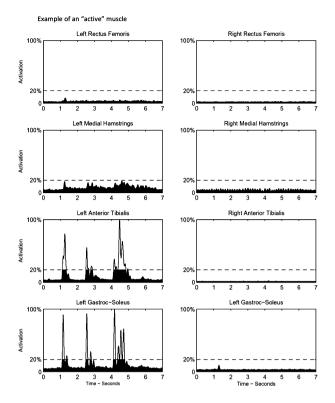


Figure 1: Muscles were designated as active when their EMG amplitude reached >20% of their maximum amplitude for all activities. The left anterior tibialis and gastrocnemius are active in this trial during the quick stretch of the left gastrocnemius in a participant with spasticity.

Design: A prospective study design was used and Institutional Review Board approval was obtained.

Participants and setting: Participants were clinically identified prospectively by the investigators through our pediatric rehabilitation clinics and center for gait and motion analysis as having predominantly spastic or predominantly dystonic hypertonia. Control participants were typically developing children with no neurologic deficits, recruited from our hospital staff member contacts.

Methods: Eight control participants, 6 participants with predominantly dystonia, and 6 participants with predominantly spasticity were recruited, ages 6 to 25 years. Surface EMG sensors were applied over 4 muscle groups of each leg. EMG and videotape recordings were obtained during rest, quick stretch, and active movement (movement initiation activities). The number of muscles active during 3 resting, 4 quick stretch, and 8 movement initiation items were averaged and compared across subject groups. All participants had some low level of background muscle activity. Because of this background activity, muscles were considered active when their EMG amplitude reached 20% of the maximum recorded amplitude for that muscle during all test activities.

Results: Control participants showed minimal spread of muscle activity during resting, quick stretch, or movement initiation activities. Participants with predominantly spasticity showed multiple muscle responses and increased amplitude of response to quick stretch but not to movement initiation activities. Participants with predominantly dystonia showed

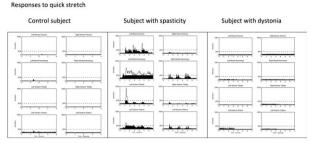


Figure 2: EMG responses of the 8 recorded muscles when the left rectus femoris was stretched rapidly. The participant with spasticity showed EMG amplitudes >20% of the maximum recorded amplitude and showed overflow to more than the stretched muscle.

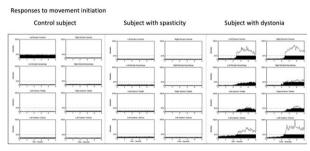


Figure 3: EMG responses of the 8 muscles recorded when the participant begins talking. The participant with dystonia showed EMG amplitudes >20% of the maximum recorded amplitude and showed overflow into multiple muscles.

Mean number of "active" muscles responding during resting activities, quick stretch activities, and movement initiation activities

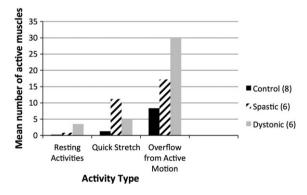


Figure 4: Mean number of active muscles responding in each subject group during resting activities, quick stretch activities, and movement initiation activities.

multiple muscle responses to movement initiation activities but not to quick stretch. A one-way ANOVA indicated significant differences between the three groups for activities at rest

Distribution of muscle responses by subject group

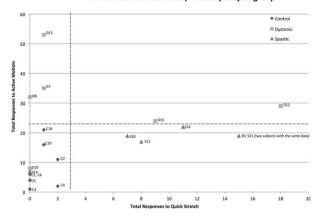


Figure 5: Distribution of muscle responses by subject group.

(p=0.055), quick stretch (p=0.007), and movement initiation (p=0.002).

Conclusions: EMG data collected with this protocol can help the clinician distinguish spastic from dystonic hypertonia.

D7

The relationship of secondary dystonia and choreoathetosis with activity, participation and quality of life measures in children with dyskinetic cerebral palsy

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Background/Objectives: To date, little information is available as to how, and to what extent dystonia and choreoathetosis affect the functional activity, participation and quality of life in children with dyskinetic cerebral palsy (CP). Yet, the primary goal of therapeutic interventions is to enhance the child's ability to perform activities in the context of daily life, which are closely related with participation and quality of life (QOL). Therefore, knowledge of the impact of secondary dystonia and choreoathetosis on the different levels of the ICF model and QOL is important to guide treatment interventions. The objective of this study was to determine the relationship between dystonia and choreoathetosis and the level of activity, participation and quality of life in children with dyskinetic CP.

Design: cohort study (correlational).

Participants and setting: This study included 54 participants with dyskinetic CP (mean age 14y 6mo, SD 4y 2mo), recruited from five Belgian Flemish special education schools for children with motor disabilities.

Methods: Dystonia and choreoathetosis were measured with the Dyskinesia Impairment Scale (DIS). Activity measures included the Gross Motor Function Measurement (GMFM-88), the Functional Mobility Scale (FMS), the Jebsen-Taylor Test of Hand Function (JTT), and the Abilhand-Kids Questionnaire (ABIL-K). The Life Habits Kids (LIFE-H) was used as a measure for social participation. For qual-

ity of life, the Quality of Life Questionnaire for children with CP (CP-QOL) was applied. Spearman's rank correlation $(r_{\rm s})$ was used to assess the relationship between both motor disorders and the activity, participation and quality of life measures.

Results: Significant moderate to good correlation coefficients were found between dystonia and the activity scales with $r_{\rm s}$ varying between -0.64 and -0.71. Fair correlations coefficients were found with the LIFE-H ($r_{\rm s}$ =-0.42) and the CP-QOL ($r_{\rm s}$ =-0.32). For choreoathetosis, no or only weak relationship was found with the activity, participation and quality of life scales.

Conclusions: This study is the first to examine the relationship between dystonia and choreoathetosis in children with dyskinetic CP and their activity abilities, participation and quality of life. A strong relationship was found between the presence of dystonia and activity scales. Similarly, considerable association was found with participation and quality of life scale, although into a lesser extent. For choreoathetosis, little or no relationship was found. These findings seem to suggest that it might be most crucial to focus on dystonia reducing intervention studies.

D8

Muscle strength after botulinum neurotoxin injection in children with cerebral palsy

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Background/Objectives: Spasticity is a common problem in children with cerebral palsy (CP), which can affect motor development. Botulinum neurotoxin injection (Btx) can be used to reduce spasticity. The drug effect is maximal after 4 to 6 weeks and lasts about 3 months. Muscle weakness has been reported, but we have found only one study that has measured muscle strength (Bjornson et al 2007), showing an increase 24 weeks after injection. The reports are conflicting and it is not fully known how Btx affects voluntary muscle strength. Muscle weakness is common in children with CP and has been shown to correlate with walking ability (Eek et al 2008). The aim of the study was to follow muscle strength, before and after injection with Btx, in children with CP.

Design: Prospective cohort study.

Participants and setting: Children were recruited consecutively from the spasticity clinic at the Regional Rehabilitation Centre in Gothenburg, Sweden. Twenty-three children with spastic CP, able to walk without support, were recruited. Three were lost to follow up, resulting in 20 children included, eight girls and 12 boys, 4 to 13 years old (mean 7.7). Sixteen children had unilateral and four bilateral involvement. Btx injection was made in the gastrocnemius muscle in 24 legs.

Methods: Measurement of muscle strength in plantar flexors was made with a handheld device. Lever arm was measured with a tape measure, torque was calculated and normalized to body weight (Nm/Kg). Muscle strength was compared before Btx, at peak effect and when Btx has leveled off. Not treated muscles served as control to treated muscle groups. Gait

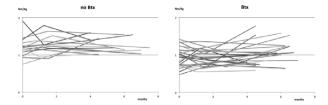


Figure 1: Muscle strength in plantar flexors, before, at peak Btx and at follow up.

velocity was registered and a 2 dimensional (2D) gait analysis was performed with registration of angle in knee and ankle at initial contact (IC) and maximum values during stance phase. Data was analyzed with independent samples *t*-test for comparison of treated/not treated leg before treatment, and paired samples *t*-test for comparison of data before and after treatment (SPSS).

Results: Before treatment, muscles subject to Btx were weaker than muscles not in question for treatment (NoBtx mean 1.21 Nm/kg (SD 0.27), Btx 0.93 (0.26), p=0.003). There were no differences at peak Btx. At follow up after treatment, there was an increase in plantar flexor strength in muscles treated with Btx (before 0.93 (0.26), follow up 1.05 (0.31), p=0.023). There was no change in gait velocity after treatment. At peak Btx the 2D gait analysis showed improvement in knee extension, both at IC (before 21.9° (9.9), peak Btx 18.5 (7.6), p=0.016) and during stance phase (before 14.0 (7.4), peak Btx 11.0 (6.6), p=0.030). There was no change in the ankle.

Conclusions: Voluntary force production in plantar flexor muscles was not decreased by Btx, on the contrary it increased at follow up. Gait pattern improved in terms of better knee extension in stance. Adequate muscle strength is important to maintain the ability to walk, and knowledge of how treatments affect muscle strength is necessary when selecting interventions, not to make muscles weaker with treatment.

D9

High passive stresses in spastic muscle are not generated from myofibrils for children with cerebral palsy

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Background/Objectives: For children with cerebral palsy (CP), high passive stresses generated in spastic muscles have been observed at many structural levels including: whole muscle, bundles of muscle fascicles, bundles of fibres, and single fibres. The origins of these high passive stresses have been attributed to both increased collagen content in the extracellular matrix and to the giant intra-sarcomeric protein titin. While the evidence at the higher structural levels of organization is well accepted, there is no data for high passive stresses at the level of the myofibril [1,2]. The purpose of this work was to investigate the generation of passive stresses within single myofibrils

as a function of sarcomere length from spastic muscle (CP participants) compared to non-spastic (non-CP participants) samples.

Design: Cross-sectional study.

Participants and setting: Muscle biopsies from adductor longus and gracilis were obtained from patients with spastic quadriplegia, GMFCS levels III-V; n=4 and from non-CP controls (n=2) during the course of tendon release procedures for the treatment of hip displacement and Perthes or developmental hip dysplasia, respectively.

Methods: The passive force generation of individual myofibrils from spastic muscle samples was measured using a single myofibril apparatus which allows for measurement of myofibril force production with an accuracy of <1nN. Myofibrils were lengthened from a mean SL of $2.0\mu m$ ('slack' length) to mean SL's of 2.4, 2.8, 3.2, 3.6 and $4.0\mu m$. At each target length, the myofibril was held for 1 minute to allow for stress relaxation before the passive stress generated was recorded. The sample was then returned to slack length for 4 minutes before repeating the test at the next SL target length.

Results: 36 individual myofibrils from CP muscle biopsies were tested along with 8 myofibrils from non-CP muscle biopsies. Figure 1 shows that for both spastic and non-spastic muscle, no differences in stresses at similar sarcomere lengths were observed.

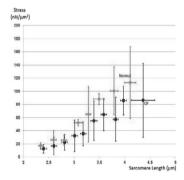


Figure 1: Stress versus sarcomere length for CP.

Conclusions: Despite the presence of underlying muscle spasticity, we did not see a difference between the passive stresses generated per sarcomere length for matched myofibril samples from participants with CP as compared to non-CP participants. As such, muscle stiffness observed clinically in patients with CP does not seem to be related to increased passive force generation at the myofibrillar level. While the sarcomeres in CP do appear normal, it has been shown previously that they tend to be abnormally long. Elongation of these sarcomeres may allow for an increased tension to develop in titin (with a subsequent increase in myofibrillar stress generation). More likely, however, the results of this study suggest that increased stiffness in spastic muscle is derived from other sources (e.g. the extracellular matrix) with little or no influence from the myofibril/sarcomere.

References:

- 1. Smith et al. 2011 J. Physiol 589: pp. 2625-2639.
- 2. Friden and Lieber 2003 Muscle and Nerve 26: pp. 157–164.

D10

Mitochondrial enzyme activity is reduced in skeletal muscle in children with cerebral palsy

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Background/Objectives: Compared to typically developing (TD) children, the energetic cost of movement is increased and endurance capacity is reduced in children with cerebral palsy (CP). These impairments have been primarily attributed to inefficient muscle activation patterns and cardiorespiratory factors. However these changes could also be due to reduced oxidative capacity (i.e., mitochondrial function) in skeletal muscle. The purpose of this study was to evaluate the maximal activity of various mitochondrial enzymes in skeletal muscle of CP and TD children to potentially understand the mechanisms behind their reduced endurance capacity.

Design: Cross-sectional study.

Participants and setting: Under a protocol approved by the Institutional Review Board of the University of California, San Diego, 13 participants (CP, n=7, 12.6 [5.6]y-old, GMFCS I [n=2], GMFCS II [n=4], GMFCS IV [n=1]; TD, n=6, 16.4 [1.3]y old) were recruited. Parental and age-appropriate consent was obtained from all the participants. Children with CP were undergoing hamstring lengthening while TD children were undergoing anterior cruciate ligament reconstruction.

Methods: Gracilis muscle biopsies were flash frozen in liquid nitrogen-cooled isopentane and stored at -80°C. Approximately 50mg of muscle was used to assess the maximal activity of mitochondrial enzymes: Complex I (CI; NADH:ubiquinone oxidoreductase), Complex III (CIII; ubiquinol cytochrome c oxidoreductase), Complex I+III (CI+III; NADH cytochrome c

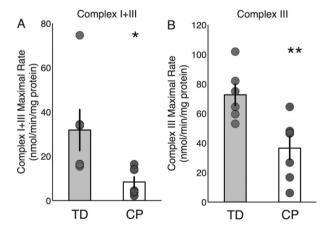


Figure 1: Maximal activity rates of mitochondrial complex I+III (A) and complex III (B) are significantly reduced by $\sim 50\%$ & $\sim 75\%$ respectively in children with Cerebral Palsy (CP) compared to children with Typical Development (TD). The bar graphs indicate mean \pm SEM, the asterisks indicates statistically significant effect between groups (*p<0.05, **p<0.01).

oxidoreductase) and citrate synthase (CS) using standardized protocols.

Results: Maximal activities of CIII (37 [8] vs 73 [7]nmol/min/mg protein) and CI+III (8 [2] vs 32 [9]nmol/min/mg protein) were reduced by 50 and 75% (p<0.05), respectively, in CP versus TD (Figure 1A,B). Preliminary data suggests that there were no differences between groups in the maximal activities of CS or CI (n=3/group).

Conclusions: These results demonstrate that the maximal activity of components of the electron transport chain are impaired in children with CP. Specifically, this impairment appears to be localized to complex III of the electron transport chain, and further studies will focus on potential mechanisms underlying this difference. Taken together, these results may be relevant to the increased fatigability and reduced endurance capacity seen during movement in children with CP and could potentially open up new avenues of therapeutic intervention.

Free Papers E

E1

The effects of Kinesio Taping® on activity and participation in children with unilateral spastic cerebral palsy: two blind-randomized control trials

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Background/Objectives: New alternative approaches used in rehabilitation process are promising to facilitate motor learning for instance neuromuscular taping. Kinesio Taping® (KT) is one of the most widely used techniques on account of its nature including elasticity, adhesive, latex free, thin and can be stretched in longitudinal plane. There are few studies showed effects of KT in children with CP and results are conflicting. However, no randomized control trial has been showed impression of KT on both upper and lower extremities. This study was to investigate the effects of KT application on activity and participation in children with Unilateral Spastic CP.

Design: Two-blind randomized controlled trial.

Participants and setting: Thirty children with unilateral spastic CP completed this study. The trial comprised a 12-week intervention period. Participants were assessed before treatment, and after 12 weeks of treatment. The inclusion criteria were (1) the children with CP aged between 7 and 14 years. The exclusion criteria were any orthopedic surgery or Botulinum neurotoxin injection in the past 6 months and showing allergic reactions to the adhesive compound of KT.

Methods: All participants were evaluated with The Functional Independence Measure for Children (WeeFIM), Bruininks-Oseretsky Test of Motor Proficiency (BOTMP) and Gross Motor Function Measurement (GMFM D&E), short-term muscle power test (Muscle Power Sprint Test), agility test ($10 \times 5 \, \mathrm{m}$ sprint test), functional muscle strength test ($30 \, \mathrm{s}$ Repetition Maximum test). Two research assistants, certified in KT courses, applied taping. Statistical Difference in pre and post treatment was analyzed with Wilcoxon signed-rank test for

each group. Mann-Whitney U test was used for comparing the improvement differences between KT and control group. Level of significance was accepted as p<0.05.

Results: KT group had significant improvement in gross motor function D&E and BOTMP gross, functional muscle strength (Lateral step-up test right, Lateral step-up test left and sit to stand) and agility (respectively; Z=-2.201, -2.812, -2.240, -2.560, -2.632, -2.295, -2.840, p<-0.05). Compared to participant restriction assessments, also KT group had significant differences in WeeFIM total, self-care and mobility scores (respectively, -3.315, -2.437, -2.232, p<-0.05). When the groups were compared for differences between pre-post treatment, there were significant differences in 10.5m sprint test, lateral step-up test right, sit to stand, attain stand through half kneel right, BOTMP Gross dimension, WeeFIM total and Self-care domains (respectively, U=37.500, 56.500, 56.000, 42.000, 49.000, p<-0.05).

Conclusions: In daily life activities, there are few of alternative invasive applications to support physiotherapy and rehabilitation. KT is promising additional approach to increase proprioceptive feedbacks and improve activity limitation and participation restriction.

E2

Move it to improve it: using a web-based therapy program to increase physical activity in children and adolescents with congenital hemiplegia

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Background/Objectives: Move it to improve it (Mitii TM) is a web-based therapy program which combines upper-limb and cognitive training with physical activity. This study examines whether Mitii TM training can increase physical activity capacity and performance in independently ambulant children with congenital hemiplegia.

Design: Randomised waitlist controlled trial.

Participants and setting: Children and adolescents with congenital hemiplegia (n=82; 42 males; age 11y 6mo [2y 5mo]) classified at GMFCS levels I=39 and II=43 completed MitiiTM in their home environments.

Methods: Participants were matched in pairs and randomly allocated to receive 20 weeks of 20 to 30 minutes of daily MitiiTM training (Mitii), or waitlisted receiving usual care (Control). Each MitiiTM program comprised 40% gross-motor activities, including sit-to-stands, squats, lunges, and balance exercises. Physical activity was assessed using the MobQues28, 6 minute walk test (6MWT), functional strength tests (maximal repetitions of sit to stand, lateral step up, half-kneel to stand in 30-s), and 4 day ActiGraph®GT3X+ accelerometer records at baseline and 20 weeks. Independent t-tests and linear regression were calculated by intention to treat with last observation carried forward. Data are mean ± 1 SD, α =0.05.

Results: Groups were similar on all characteristics at baseline (p>0.05). At 20 week, 38 (93%) participants in Mitii and 35 (85%) participants remained in Control. Mitii participants completed 28.0 (14.6) hours of MitiiTM training (60% potential dose; range 3.6-54.5h) on 72 (34) days. Control participants received Physiotherapy monthly (n=6), once (n=8) or not at all (n=21), total dose 1.3 (1.4) hours. Mitii participants demonstrated significant improvements in functional strength (19 combined reps, 95% CI=10-28, p<0.001) and 6MWT (33m, 95% CI=14-52, p=0.001) compared to Control at 20 weeks. There were no differences in MobQues28 scores (-4 points, 95% CI=-8 to 1, p=0.13) or physical activity (6 counts/min, 95% CI=-87 to 100, p=0.89). Results were consistent across GMFCS levels. Mitii™ training dose did not predict the size of change demonstrated in functional strength (R^2 =0.03, p=0.40), MobQues28 (R^2 =0.30, p=0.37), or physical activity counts/minute $(R^2=0.28, p=0.22)$, but 6MWT demonstrated a trend $(R^2=0.28, p=0.07)$. MitiiTM group allocation significantly predicted improvements in functional strength (R^2 =0.41, p<0.001) and 6MWT (R^2 =0.20, p=0.001), but not MobQues28 (R^2 =0.27, p=0.16), or physical activity counts/minute ($R^2=0.17$, p=0.96). Conclusions: MitiiTM training can improve physical activity capacity though not performance in independently ambulant children with congenital hemiplegia. There was no change in activity limitations though this may reflect ceiling effects with the MobQues28. This study suggests that the web-based training program MitiiTM can be used to train activity capacity but interventions targeting an increase in habitual physical activity may need to attend to environmental and motivational aspects of behavior change.

E3

The effect of participation in an intensive adaptive sports program on walking function and endurance in children with cerebral palsy

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Background/Objectives: It is important that children with cerebral palsy (CP) maintain a high level of physical fitness similar to typically developing children to avoid the natural functional decline that occurs with age in this population. By participating in adaptive sports programs developed specifically for children with CP, individuals are given an opportunity to be physically active which may improve their level of fitness and functional ability. The purpose of this study was to determine the effect of participation in adaptive sports programs on walking function and endurance.

Design: This was a retrospective analysis of children with CP ages 6 to 20 years who participated in a local adaptive sports program from spring 2004 to summer 2012. There were 519 participants in the programs. Of these, 326 participants (mean age 11.5 [3.2] years) had pre and post data recorded. The 326 participants consisted of a total of 109 children, many of whom attended multiple programs throughout the years.

Participants and setting: Children attended programs in the spring, summer and fall. Summer programs (n=269) were held 5 days/week for a total of 4 weeks. Spring and fall basketball programs (n=57) were held 1 night/week for 8 weeks.

Methods: Outcome measures: Timed Up and Go (TUG), modified 6-minute walk (m6MW, children were allowed to run), and 25-foot walk/run. Data Analysis: Data was analyzed separately for summer and spring/fall programs. A repeated measures ANOVA, using Gross Motor Function Classification System (GMFCS) as a between participants factor was conducted. A longitudinal analysis was conducted with time as a level 1 predictor and GMFCS as a level 2 predictor.

Results: Summer: There was a significant main effect for the TUG (p=0.025) and m6MW (p<0.001) but no significant change in 25-foot walk/run. Spring/Fall basketball: There was no significant change in any of the outcome measures. Longitudinal data showed a significant improvement (*p*=0.045) in the TUG, m6MW and 25-ft walk/run over time. GMFCS level III made the most significant gains.

Conclusions: Walking function and endurance improved as a result of attending intensive adaptive sports programs. The less intensive basketball programs did not show significant gains. Higher frequency of program attendance showed improvements in the TUG, m6MW and 25-ft walk/run over time. Participation in intensive adaptive sports programs appears to be beneficial for children with CP, especially those who attend multiple programs over time.

E4

Improving the participation of youth with physical disabilities in community activities: an evaluation

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Background/Objectives: Youth with physical disabilities experience restrictions to participation in community-based leisure activities, which can lead to poor health outcomes. There is little evidence; however, about how to promote adolescents' involvement in these activities. The purpose of this study was to examine whether an intervention that aims to remove environmental barriers and provide education for parents, in the form of coaching, can effectively improve youth's participation in their community.

Design: An Interrupted Time Series design was employed where replication of the intervention effect was examined across three individualized participation goals and across participants. Participants and setting: Six adolescents (5 males, 1 female), ages 13 to 19 years (mean=16.33, SD=2.4) with physical disabilities and living in Ontario participated in a 12-week environmentfocused intervention. An occupational therapist worked with each youth and their family individually to set 3 participation goals and identify and implement strategies to remove environmental barriers. The therapist also coached the youth and parents about methods to overcome barriers autonomously. Methods: Using a multi-baseline design, interventions for each

goal were introduced at different time points. The Canadian

Occupational Performance Measure (COPM) was used to set

and monitor participation goals; the performance of each goal was repeatedly measured over a course of 20 weeks at baseline, intervention and follow-up. For each goal, a series of data points that represents goal performance was plotted and analyzed using visual inspection and the celeration line approach; the latter detects change in trend and slope across baseline and intervention phases.

Results: Improvements in COPM performance scores were clinically significant (minimum 2-point improvement) for 83% of the participation goals (15 out of 18 goals); an average change of 4.5 points in the performance scale (SD=2.6) was observed within the first 2 weeks following the intervention. Statistical analysis using the celeration line demonstrated that the proportion of data points falling above the line increased in the intervention phase for 17 out of the 18 goals, which in turn indicated a significant replication of treatment effect.

Conclusions: This study is the first to examine an intervention aimed at increasing leisure participation in this population through changing only the environment. The results indicate that environment-focused interventions are feasible and effective in promoting youth participation. Such findings can inform the design of a larger study that aims to test this intervention among a larger and more diverse sample of youth with physical disabilities and, consequently, can guide clinical practice.

E5

Effectiveness of a lifestyle program among adolescents and young adults with cerebral palsy: a randomized controlled trial

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Background/Objectives: Sufficient physical activity and fitness is considered of major benefit to a healthy lifestyle. Nevertheless, research has consistently shown that persons with cerebral palsy have low levels of physical activity and fitness. Furthermore, this unfavorable lifestyle is expected to negatively affect fatigue, participation and quality of life. To assure healthy aging, applying a lifestyle program to increase daily physical activity and fitness seems beneficial in adolescents with cerebral palsy.

Design: Multi-centre single blind randomized controlled trial. Participants and setting: Fifty-seven adolescents and young adults with spastic cerebral palsy, classified as level I-IV on the Gross Motor Functioning Classification System were recruited from 2 university hospitals and 4 rehabilitation centres in the Netherlands.

Methods: Participants were randomly assigned to either an intervention or control group. The control group received no intervention, which is regular care in the Netherlands. The intervention group followed a lifestyle program aiming to permanently increase physical activity and fitness. The intervention had a 6-month duration and consisted of individual counseling on daily physical activity and sports participation. Fitness training was offered during the first 3 months. Participants were measured before, directly after, and 6 months after

finishing the intervention. Fitness was measured by peak oxygen uptake using oximetry during a progressive protocol on a cycle or armcrank ergometer. Daily physical activity was objectively measured for a period of 3 days using accelerometry-based activity monitors. Fatigue, participation and quality of life were assessed by questionnaires. GEE-models were applied to examine longitudinal effects of the intervention.

Results: No significant effects over time were found for daily physical activity between the two groups. A significant effect, in favor of the intervention group, was found for fitness, 3 months after finishing the fitness training (p<0.01). However, this effect was no longer present 9 months after finishing fitness training. Favorable intervention effects were found on fatigue (p=0.02) during the intervention and quality of life with respect to bodily pain (p=0.01) and mental health (p=0.03) during follow up. Conclusions: The lifestyle program did not achieve a behavioral change toward increased daily physical activity. However, results of the intervention on fitness were promising as still 3 months after finishing the fitness training favorable effects were seen. Nevertheless, this effect on fitness did not last on the long term. The lifestyle intervention favorably affected secondary outcomes as fatigue and quality of life.

F6

Ambulant children with cerebral palsy participate in reduced levels of vigorous physical activity compared to their peers with typical development

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Background/Objectives: Children with cerebral palsy (CP) participate in lower levels of moderate-to-vigorous physical activity (MVPA) than their typically developing (TD) peers. Recent research suggests that vigorous physical activity (PA), and not moderate PA, is consistently associated with lower waist circumference, lower body mass index and lower systolic blood pressure in children. This suggests an emphasis should be placed on accumulating vigorous PA alone rather than MVPA. Vigorous PA includes activities such as brisk walking, running and jumping, which children usually accumulate through sport. Regular participation in sport is known to be low among children with CP, even among those with minimal impairments. Although this suggests that they may accumulate low levels of vigorous PA the difference in objectively measured vigorous PA between children with CP and TD children has not been investigated. The purpose of this study was to compare levels of sedentary, light, moderate and vigorous PA, and adherence to PA guidelines between children with CP and TD children.

Participants and setting: Data was collected on 33 children with CP (aged 8.5 [1.2]y) and 33 age- and sex-matched TD children. Children with CP were classified in Gross Motor Function Classification System level I (n=23), level II (n=6) and level III (n=4). Methods: PA was measured with an RT3 accelerometer. Children wore the RT3 for at least 3 days and for at least 10 hours per day. Time spent in sedentary, light, moderate

and vigorous PA was calculated by applying cut-points to

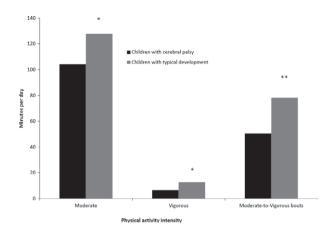


Figure 1: Time spent in moderate physical activity, vigorous physical activity and moderate-to-vigorous physical activity in 10-min bouts. *P>0.05: ** P>0.01.

accelerometer counts. Time spent in MVPA in 10-minutes bouts and time spent in sedentary activity as a percentage of total wear-time was also calculated. Finally the number of children in each group who met the PA guideline of 60 minutes MVPA daily was calculated. Data was checked for a normal distribution using the Kolmogorov–Smirnov test. Variables with a skewed distribution were square-root transformed. Independent *t*-tests and chi-square tests were used to investigate between group differences.

Results: Children with CP spent more time in sedentary behaviour than TD children (25.2 [2]% vs 17.5 [6.6]%; p<0.001). They also spent less time in moderate PA (p<0.05), vigorous PA (p<0.05) and MVPA in 10-min bouts (p<0.01) (Fig. 1). Although a slightly higher proportion of TD children met the guideline, there was no significant difference between groups (93.9% vs 97.0%).

Conclusions: Results suggest that as well as accumulating less moderate PA, children with CP accumulated less vigorous PA that their TD peers. They also accumulated less time in sustained MVPA. This may have an impact on their cardiometabolic health. Although a high proportion of children with CP attained adequate levels of PA according to the current guideline, they may have achieved this by accumulating moderate PA only. As recent research demonstrated possible protective effects of vigorous PA in this cohort, more guidance on the quantity of vigorous PA children with CP should accumulate is required.

E7

'I can participate'. Children with disabilities and participation in physical activity: a mixed methods study in a rehabilitation context

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Background/Objectives: Studies have documented that children with disabilities experience larger participation restrictions in everyday activities and physical activities, and are participating

Design: Cross-sectional study.

less in decision making, than peers without disabilities. Increased participation in activities is assumed to be beneficial physically, psychologically and socially for children with disabilities. A certain level of participation represents a prerequisite for learning, activity performance, development of friendship, and satisfaction in everyday life. This study investigates preferences, actual participation and enjoyment in physical out-of-school activities in children with physical disabilities. *Design:* This presentation is based on the baseline data of a longitudinal study with mixed methods design.

Participants and setting: Data are based on structured and qualitative interviews of 298 children, 45% girls, 6 to 17 years of age, attending a 3 weeks long intensive rehabilitation program at Beitostølen Healthsports Center (BHC) in Norway.

Methods: The Canadian Occupational Performance Measure (COPM), Children's Assessment of Participation and Enjoyment (CAPE) and Preferences for Activities of Children (PAC) scales were used for gathering data of relevance at the start of their rehabilitation stay. In addition qualitative interviews were performed with a strategic sample of ten children and their parents.

Results: The study reveals a relatively high level of participation in physical activity, but the children want to be even more active than they actually are. They have clear preferences for activities, they show ability to express good reasons for these preferences, and their joy of being active outweighs their effort. Children are most frequently active with family members, but would prefer to be more active with peers. There are more similarities than differences between boys and girls and between age-groups on the parameters investigated in this study. Knowledge gained through this study points out the importance of letting the children learn a few self-initiated activities well enough to be performed with peers.

Conclusions: This means that the rehabilitation field should focus on self-determination, competence, resources, preferences, and possibilities of participation with peers. By this, children will experience more meaningful participation.

E8

Results of a 3-year prospective cohort study investigating the influence of home-based therapy on patients with cerebral palsy in GMFCS levels IV and V

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Background/Objectives: Severely affected cerebral palsy (CP) patients (GMFCS 4 and 5) may struggle to respond to rehabilitative programs subsequent to infrequency and poor co-operation. This population may benefit from a high frequency home-based program. Thus, the feasibility of a high frequency home-based therapy tailored to GMFCS type 4 and 5 patients was explored. Design: Studies of Therapy: Non randomized prospective cohort treatment study.

Participants and setting: To date, 367 American and South American cerebral palsy type 4 and 5 patients participated. Inclusion criteria was limited to patients over 5 years old, not

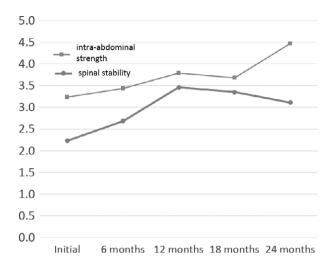


Figure 1: Measures of intra-abdominal strength and spinal stability over time.

having undergone surgery over the course of the study, and having at least a 3 year follow up. This left 100 patients haven taken part in the study.

Methods: At baseline and every 6 months, parents of CP patients were provided a 3 day lesson by experienced physical therapists teaching a safe and simple home-based manual therapy with focus to provide cyclic motion strain to the weaken midsection of their child. Stimulation guidelines (pressure magnitude and frequency) were instructed and monitored for consistency using a custom force gauge integrated into the therapy. Therapy was encouraged for at least 30 minutes 5 times a week. CPCHILD questionnaires were collected at baseline and bi-annually for 3 years by the physical therapists. Additionally, measures of intra-abdominal strength (force for 1 inch of abdominal compression), seated spinal stability (scored according to minimal support required for 10s of independent sitting), and the GMFCS classification were taken initially and bi-annually for 2 years. Study directives were approved by independent ethical committees. All follow up measures were blinded from preceding data. Non-parametric Wilcoxon tests were utilized to perform data analyses.

Results: Baseline CPCHILD scores agreed with published mean values per GMFCS class. To date, CPCHILD scores of GMFCS 4 patients improved consistently by 8% from 51.55 to 56.02 and GMFCS 5 improved by 14% from 48.74 to 55.54 after 36 months of home therapy (p<0.05). Measures of intra-abdominal strength improved from 3.23 to 4.46 lbs/inch of compression and seated spinal stability improved from 2.3 to 3.1 after 36 months (p<0.05). The improvements of intra-abdominal strength and seated spinal stability corresponded closely. The average GMFCS scores for type 5 improved to a score of 4 while types GMFCS 4 did not undergo any significant changes after 24 months.

Conclusions: Results suggest a high frequency home-based therapy for CP patients to be a feasible platform for the improved health and wellbeing of severely affected cerebral palsy patients GMFCS types 4 and 5. An independent multi-center study is now being piloted to further explore the benefits of home-based therapy for these patients.

F9

Focus on participation for children and youth with physical disabilities: a knowledge translation implementation study

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Background/Objectives: Although participation in communitybased leisure activities of children and youth with physical disabilities plays a key role in their development, health and wellbeing, it is not well integrated into practice. This knowledge translation (KT) strategy tested a 6-week intervention plan to increase clinicians' awareness and to bring about change in practice towards a focus on participation of children with disabilities.

Design: A quasi-experimental (post-test only) design was employed using reflective qualitative methods to evaluate change in practice.

Participants and setting: Two interdisciplinary groups of 14 female clinicians (e.g., occupational therapists, physiotherapists, speech language pathologists) participated in the study (50% response rate of those eligible). All work in a large rehabilitation centre in Greater Montreal with children with physical disabilities, and their years of clinical experience range from 6 months to 15 years (mean=5.7).

Methods: Clinicians participated in six learning group sessions of 2 hours each, facilitated by a Knowledge Broker. Based on self-identified learning needs around participation, research evidence was selected and implemented using a case-based learning approach. Three months post-intervention, clinicians were individually interviewed by drawing on the Professional Evaluation and Reflection on Change Tool to evaluate their change in practice behavior regarding the topic of participation. Interviews were transcribed, coded and organized using Nvivo. Two researchers independently performed thematic analysis of the data.

Results: Four themes emerged from the data, two of which illustrated the personal and professional impact of the intervention on practice. Clinicians experienced a sense of empowerment and validation of clinical wisdom and reported a change in thinking and behavior by suggesting strategies for integrating participation into their clinical routine (e.g., community involvement, setting and measuring participation-based goals). Barriers and facilitators for implementing change also emerged, including issues around feasibility and a supportive organizational mandate, respectively. The two remaining themes - Elements of the learning environment (e.g., KT for meeting informational needs) and Motivation to learn (e.g., desire to link research to practice) - were described as facilitators to knowledge uptake.

Conclusions: This KT strategy is one of the few examples of implementation studies demonstrated in pediatrics. It was effective in shifting clinicians' focus towards the notion of participation and establishing a readiness for change and, therefore, minimized the current knowledge-to-practice gap. This protocol can be used in further KT initiatives among a broader range of stakeholders such as managers and directors of rehabilitation centers, families and service providers in the

community, in order to foster a sustainable behavioral change.

E10

School-based physical therapy services for children with cerebral palsy within the United **States**

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Background/Objectives: To evaluate school-based physical therapy, reliable data on the services used is needed. We identified the current physical therapy services provided for students with cerebral palsy (CP) in school-based practice in the United States.

Design: Descriptive study.

Participants and setting: Within a larger study (PT COUNTS), physical therapists (PTs) (N=112) from four regions of the US (Northeast, Southeast, Central, Northwest) reported on school-based services provided to 101 children with CP out of 304 they followed from their caseloads, age 5 to 12 years.

Methods: PTs were trained to use the School-Physical Therapy Interventions for Pediatrics (S-PTIP) data collection system to provide detailed service information. PTs reviewed the S-PTIP manual and data form, completed online training and passed reliability testing. S-PTIP intra-rater reliability was examined; 89.6% consistency was found by comparing data of 25 live to videoed therapy sessions. PTs used the S-PTIP weekly for 6 months during the 2012 to 2013 school year to report on time spent within activities, interventions used, time spent in various service delivery methods, time spent in services on behalf of the student, where services were provided, and participation of students within therapy sessions. Descriptive statistics were calculated.

Results: PTs provided services for a mean of 31 minutes/week (SD=20.2) to students with CP. Within the 6 months (vacations excluded), 22% of the time no services were provided, primarily due to students' absences (39%) and school closures (28%). The most frequent activities (mean minutes/week) were mobility (10.7, SD=10.6), sitting/standing/transitions (9.5, SD=11.2), and physical education/recreation (7.3, SD=7.4). The least frequent was self-care (2.0, SD=2.8). The largest mean number of interventions received were neuromuscular (primarily balance and motor learning techniques) (37.7, SD=17.7), mobility (primarily stair and door training) (21.1, SD=17.0), and musculoskeletal (primarily functional strengthening) (18.0, SD=12.4). The least mean number of interventions utilized was cardiopulmonary (5.4, SD=5.2). More mean service minutes/week were delivered to the students individually (29.2, SD=21.3) than in a group (5.3, SD=6.4) and separate from school activity (19.9, SD=13.4) than within school activity (13.7, SD=17.1). Services on behalf of students occurred for a mean=14.5 minutes/week, SD=8.9. Most students received services within the school (87.1%) and students' mean participation rated on a 1 to 5 scale was 4.9, SD=0.96.

Conclusions: School-based physical therapy services for students with CP are primarily mobility, transitions and recreation movement using motor learning, balance and functional strengthening techniques, all of which have some research support. Services are limited in intensity and primarily provided individually to students and separate from school activities, all of which are challenged based on research. Given these data, evaluation of services and appropriate effectiveness studies can be conducted.

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F1

Child Apolipoprotein E gene variants and risk of cerebral palsy: estimation from case family triads

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Background/Objectives: Three alleles, $\varepsilon 2$, $\varepsilon 3$ and $\varepsilon 4$ of the gene APOE encoding for Apolipoprotein E (ApoE) have been described, resulting in corresponding differences in protein structure. Presence of the &4 allele is associated with Alzheimer disease and prolonged recovery after traumatic brain injury. However, regarding the etiology of CP, APOE association studies have been inconclusive. Case-parent triad data are considered a robust basis for studying association between variants of a gene and a disease. In previous studies we found that presence of the &4 allele, as well as presence of the T allele of the rs59007384 SNP in the TOMM40 gene regulating the production of ApoE were associated with more severe manifestation of cerebral palsy (CP). The aim of this study was therefore to use case-parent triad data to investigate if CP is associated with the APOE&4 allele, the T allele of the rs59007384 SNP or with the haplotype of APOEε4 and rs59007384 T.

Design: Case-parent triad study.

Participants and setting: We included 255 children recorded in the CP Register of Norway and their parents. Genotyping was performed on DNA from buccal epithelial cells. In 215 families DNA was available for all family members.

Methods: We estimated relative risks (RR) with 95% confidence intervals (CI) of child APOE genotypes, rs59007384 variants and combined haplotypes compared to population average risk using Haplin, a statistical software designed to study multimarker associations among case triads. This software assesses if the distribution of haplotypes among the children with CP deviates from what would be predicted by Mendelian inheritance of their parents' haplotypes. Estimation of haplotypes and relative risks are done with a maximum likelihood approach which includes triads with missing data.

Results: Children carrying the APOE ϵ 3 allele had a RR of 7.5 (CI: 0.99–53.7) for heterozygotes and 10.3 (CI: 1.4–79.6) for homozygotes. For rs59007384 alone we found no association with CP. When we studied haplotypes, the haplotype of APOE ϵ 3 and rs59007384 G was the only to give increased risk of CP (RR=2.4; CI: 1–5.7 for heterozygotes and RR=3.7; CI: 1.4–9.5 for homozygotes).

Conclusions: In contrast to our previous findings regarding severity of CP, we found a strong association between CP and the APOE ϵ 3 allele. The effect was dose-dependent. In addition we found an association between CP and the haplotype APOE ϵ 3 and rs59007384 G. This is the first time these associations have been described, and further studies are needed to confirm the findings.

F2

Clinical and neuroimaging findings in children with cerebral palsy associated with congenital cytomegalovirus

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Background/Objectives: Cytomegalovirus is a common congenital infection. It is known to cause long term neurological impairment in approximately 10% of infected infants who show signs of infection at birth, and a further 10–15% of asymptomatic infection. Cerebral palsy (CP) a known sequelae of congenital cytomegalovirus infection (cCMV). Objective: To describe the clinico-radiological profile of children with CP and confirmed or probable cCMV infection.

Design: Retrospective case series.

Participants and setting: The study cohort comprised children with CP and cCMV (n=16). CP cases reported to the Australian Cerebral Palsy Register and born in the states of South Australia or Victoria, 1993 to 2006 were included if they had (i) cCMV listed in the 'known cause' field of the Register and met the criteria for confirmed or probable cCMV infection and (ii) an available magnetic resonance imaging and/or computerized tomography report.

Methods: Neuroimaging findings and clinical details including CP type, functional mobility and associated impairments were tabulated and described.

Results: The majority of children had spastic quadriplegia (n=10) and were classified as Gross Motor Function Classification System (GMFCS) level IV or V (n=11). Associated co-morbidities confirmed at 5 years of age included epilepsy (n=10), intellectual impairment (n=15), reliance on non-verbal communication (n=10), bilateral deafness (n=9) and functional

blindness (n=2). Fifteen children had abnormal neuroimaging findings. White matter injury (n=9) and ventricular dilatation (n=11) were noted in the majority of cases. Malformations were reported in all but one case and included disorders of neuronal migration (lissencephaly, pachygyria and polymicrogyria) and other cortical malformations. Cerebellar hypoplasia was another common feature (n=6) and included hemi-cerebellar hypoplasia. Calcifications were also reported for the majority of children (n=10).

Conclusions: This study expands our knowledge of neuroimaging findings in children with CP with characteristic cCMV infection and supports the premise that CP as a sequelae of cCMV may result in severe disability. We identified some distinct patterns of neuroimaging here including brain malformations, cerebellar hypoplasia, white matter injury and ventricular dilatation that can be used to better understand the profile of CP and cCMV especially when testing for cCMV occurs after the neononatal period. Early identification of children at risk of CP associated with cCMV infection provides opportunity for referral to early intervention and secondary prevention programs. Further, with new strategies emerging to reduce the severity of outcomes from cCMV, strategies for early identification of cCMV and CP are essential. Future research with a prospective study design or case-control series of children with CP plus and minus cCMV would further support our understanding of this group of children.

F3

Causal paths to cerebral palsy in term and late preterm singletons that include fetal growth restriction

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Background/Objectives: Fetal growth restriction (FGR) and cerebral palsy (CP) are associated, but the mechanisms underlying this association are poorly understood. This analysis seeks to identify causes of FGR associated with CP in term and late preterm singletons.

Design: Population based case control study.

Participants and setting: Singletons born ≥35 weeks gestation were selected from the Western Australian case control study of CP and perinatal death in which data was extracted from pregnancy, labour and delivery records of (i) all children born 1980 to 1995 registered with moderate or severe CP not of post neonatal origin, (ii) neonatally surviving controls not so registered matched for gestation and birth year and (iii) representative samples of perinatal deaths born in the same years, providing 494 CP cases, 508 controls, 100 neonatal deaths and 73 intrapartum stillbirths for these analyses

Methods: Fetal growth was considered severely restricted (FGR) if proportion of optimal birthweight¹ (<-2 SD below median of an optimally grown population) or neonatally diagnosed growth restricted. Birth defects diagnosed by age 6 years were obtained from the State birth defects register. We examined associations between antecedents of FGR (maternal smoking, pregnancy induced hypertension (PIH) and birth defects) and outcomes stratified neonatal condition. Associations with additional factors that might suggest causal mechanisms were sought as required. Frequencies, proportions and odds ratios obtained by conditional logistic regression were calculated using SAS software

Results: 5.3% (27/508) controls met FGR criteria and 16.9% (82/485) CP. 55% FGR CP showed no neonatal neurologically abnormality, 27% encephalopathy not considered asphyxial and in 18.% encephalopathy considered on varying evidence to be asphyxial. Maternal smoking was not associated with FGR in controls (OR=0.8 (95% CI 0.2, 2.4) but was in CP. OR for CP with smoking+FGR=8.7 (2.5, 10), partially attributable to major (often cerebral) birth defects: a role for maternal recreational drug use is hypothesized. PIH was a relatively benign cause of FGR: OR for CP in the presence of FGR without PIH=4.5 (95% CI 2.6, 7.9) and with PIH=2.0 (0.7, 5.9). Major birth defects, particularly of genetic origin, were seen in 43/82 (52%) of all FGR CP ((33% of FGR CP considered asphyxial) and 22/30 (73%) FGR neonatal deaths but only 1/27 (3.7%) FGR controls. High birth length for weight suggesting recent onset FGR and associated with PIH, was observed in 7/16 FGR intrapartum stillbirths and 7/15 FGR CP considered asphyxial compared with controls (1/27) Conclusions: CP was strongly associated with FGR with major birth defects. Birth asphyxia is an intermediate factor in <20% FGR CP. Objective methods of ascertaining maternal recreational drug use are required to assess their contribution to CP aetiology. Several causal paths to CP include FGR, each making small a contribution. 1.Blair et al. BMC Pediatrics 2005.

F4

Hypertension, proteinuria, fetal growth restriction and cerebral palsy in term births

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Background/Objectives: Fetal growth restriction (FGR) is associated with cerebral palsy (CP). Pregnancy induced hypertension (PIH), particularly with proteinuria, is considered a frequent cause of FGR, suggesting the causal path PIH to FGR to CP. Estimating the risk of CP with PIH is complicated as it is an indication for early deliver; is control for gestation appropriate? Understanding the role of FGR is complicated by varying FGR definitions and small samples. We seek to clarify relationships between PIH, proteinuria, FGR and CP in term born singletons, which contribute 62% of moderate and severe congenital CP.

Design: Population based case control study.

Participants and setting: Singletons born ≥37 weeks gestation were selected from the Western Australian case control study of CP and perinatal death which extracted data from pregnancy, labour and delivery records of (i) all births 1980 to 1995 later registered as moderate or severe CP not of post neonatal origin, (ii) neonatally surviving controls not CP registered matched for gestation and birth year and (iii) representative samples of perinatal deaths born in the same years; giving 460 CP cases, 468 control and 137 perinatal deaths *Methods*: Proteinuria was considered present if noted in medical record, PIH if BP >140/>90 or SBP rise of 20mm or DBP rise of 15mm during pregnancy; severe FGR if proportion of optimal birth weight(POBW)¹<77.3% (<-2 SD of optimally grown population) or neonatally diagnosed as growth restricted; moderate FGR if 77.3% 85%. Uni and multivariate analysis used SAS software: attributable proportions estimated as: ((OR-1)×p)/(1+(OR-1)×p), OR=odds ratio, p=proportion of controls exposed

Results: 9% of term born CP was attributed to severe FGR and 2.8% to moderate FGR. Proteinuria status was unknown for ~23% of controls and CP and 7% of perinatal deaths with PIH: of the others 56.4%, 50.0% and 60.0% of controls, CP and deaths with PIH reported proteinuria. Controlling for gestation and birth year, the higher risk of CP associated with PIH was not increased by proteinuria. Conversely proteinuria increased the risk of perinatal death associated with PIH. Most PIH was not associated with severe FGR: 5.9%, 10.8% and 25.9% in PIH controls, CP and deaths respectively and most severe FGR not associated with PIH, 14.3%, 11.8% and 25% in severe FGR controls, CP and deaths. Multivariate analysis estimated that PIH in AGA births contributed 6.8% to term born CP compared with 1.8% for PIH with severe FGR and 11.9% for normotensive severe FGR

Conclusions: Proteinuria increases risk of perinatal death with PIH but not of CP. Considering only pre eclampsia underestimates the contribution of PIH to CP. PIH in AGA births contributes significantly more term CP than does PIH with severe FGR despite the its higher relative risk. Importance to public health depends on population frequency of an exposure as well as risk ratio. Normotensive severe FGR contributes 6 times as much term CP as PIH severe FGR. PIH is responsible for little of the association between CP and FGR in term births.

F5

Neurodevelopmental outcomes in a phase I pilot trial of erythropoietin and hypothermia for neonatal encephalopathy

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Background/Objectives: Hypoxic-ischemic encephalopathy (HIE) is an important cause of death and neurodevelopmental disability. Trials of therapeutic hypothermia (HT) for HIE still demonstrate rates of death or moderate to severe impairment of 45% to 55%. Erythropoietin (Epo) may be neuroprotective in HIE. The long-term safety of Epo in neonates undergoing HT for HIE is unknown.

Design: Blinded phase I open label, dose escalation study. Participants and setting: We enrolled 24 newborns undergoing HT for HIE at 5 academic medical centers in the United States. Methods: In the trial, patients received up to 6 doses of Epo. All infants underwent neonatal brain MRI that was reviewed

by a single neuroradiologist. Children were then followed after discharge in child neurology or high risk infant follow up programs per each site's clinical practice. Moderate to severe neurodevelopmental disability was defined as cerebral palsy with Gross Motor Function Classification Score (GMFCS) 3 to 5, or cognitive impairment based on Bayley Scales of Infant Development II MDI or Bayley III cognitive composite score.

Results: 22 of 24 infants (92%) were followed for over 6 months and are included in our follow up cohort. These infants were followed to mean age 23 months (SD 7.4). There were no deaths. Moderate to severe disability was present in one child (4.5%) who had cerebral palsy and GMFCS of 3. An additional 5 patients exhibited mild neurodevelopmental abnormalities. All 11 of 22 (50%) patients with a normal brain MRI had a normal outcome. Eight patients (36%) had moderate to severe brain injury on MRI, including the single patient with moderate to severe disability who suffered basal ganglia injury. Seven patients with moderate to severe watershed distribution injury exhibited the following outcomes: normal (3), mild language delay (2), mild hemiplegic CP (1) and epilepsy (1).

Conclusions: Epo appears to be safe when administered with therapeutic hypothermia for HIE. Although infants enrolled in this trial exhibited a relatively low rate of moderate to severe neurodevelopmental disability and cerebral palsy, even in the face of moderate to severe injury on MRI, further studies are needed to assess long-term safety and efficacy of this novel therapy.

F6

Trends in period prevalence of cerebral palsy, 1993–2010

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Background/Objectives: Cerebral palsy (CP) is the most common motor disorder in childhood, often impacting daily functioning and requiring extensive, lifelong supports. Understanding trends in CP prevalence can provide empiric evidence to further policy and plan for resource needs. Recent reports from international monitoring programs have described decreases in CP birth prevalence while results from U.S. survey data report stability in CP prevalence among 3 to 17 year-olds. The objective of this study is to examine trends in CP prevalence over the past 18 years in a heterogeneous, U.S. metropolitan area.

Design: Population-based, cross-sectional CP surveillance program.

Participants and setting: Through abstraction and clinician review of education and health records, the Metropolitan Atlanta Developmental Disabilities Surveillance Program (MADDSP) identified 1327 children with CP across nine surveillance years from 1993 through 2010. Children were included if they resided in the 5-county metropolitan Atlanta,

Georgia area and were 8 years old during the surveillance year and met the CP case definition.

Methods: Period prevalence was calculated as the number of children with CP for each surveillance year divided by population estimates for 8-year-olds in metro Atlanta for the same year. Average annual prevalence was calculated as the sum of CP cases from 1993 to 2010 divided by the sum of population estimates for this period. Negative binominal regression was used to model linear trends in prevalence. Chi-square Goodness of Fit test results were used to examine the fit of each trend (p>0.05). Trends were examined by sex, race/ethnicity, CP subtype and the presence of co-occurring autism spectrum disorder (ASD), intellectual disability (ID), hearing loss (HL), or vision impairment (VI).

Results: The average annual period prevalence was 3.6 per 1000 or 1 in 278 children with CP from 1993 to 2010. The overall trend in CP prevalence was stable over this time period [(0.5%, 95% confidence interval (CI) (-0.5, 1.5)]. Across all surveillance years combined, CP prevalence was significantly higher among Black non-Hispanic children (3.8) compared with White non-Hispanic (3.4, p<0.04) and Hispanic (2.8, p<0.004) children as well as among males (4.0) compared with females (3.2, p<0.001). Spastic CP was the most common subtype (79.1%) across all years; predominantly bilateral spastic CP (67.1%). ID was the most common co-occurring condition (48.6%) followed by VI (14.4%), ASD (8%), and HL (4%). Trends in CP prevalence in each sex, race/ethnicity, CP subtype and co-occurring DDs were stable from 1993 to 2010. Conclusions: While evidence of stability of CP prevalence,

despite improvements in neonatal survival, is encouraging, the lack of decreases in CP prevalence across all subgroups supports the need to continue to monitor trends as well as to accelerate the pace of etiologic and risk factor research to identify areas for prevention.

F7

Declining trends in the proportion of 8 year-old children with cerebral palsy born low birth weight, with evidence of increasing racial disparity, United States, 2002-2008

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Background/Objectives: The United States Department of Health and Human Services' Healthy People 2020 (HP2020) sets empirically derived goals for improvement of population health. The HP2020 objective for cerebral palsy (CP) is to reduce the percentage of children with CP born low birth weight (LBW, <2500 grams) from the 2006 estimated baseline of 50% to 45% by the year 2020. The purpose of this study is to evaluate initial progress toward this objective by describing trends during the period 2002 to 2008 in the percentage of 8 year-old children with CP who were born LBW based on CP surveillance data for the United States.

Design: Population-based, cross-sectional samples of children with CP.

Participants and setting: The Autism and Developmental Disabilities Monitoring Network conducted surveillance of CP among 8 year-old children in selected counties of Alabama, Georgia, and Wisconsin in 2002, 2004, 2006, and 2008 and of Missouri in 2006 and 2008. After clinician review of abstracted records, 1571 children were identified as having CP among 472 619 children who turned 8 years old during one of the surveillance years. During the 6 year surveillance period, the prevalence of CP declined from 3.6 (95% confidence interval [CI] 3.3-4.0) per 1000 in 2002 to 3.1 (95% CI 2.8-3.4) per 1000 in 2008. For the present study, birth weight information was available through linkage with birth certificates for the 1201 (76.4%) of children who were born in the same state as their residence at age 8.

Methods: Trends from 2002 to 2008 in the frequency of LBW among children with CP with available birth weight information are reported for the overall sample and stratified by race/ ethnicity. Logistic regression was used to test the significance of trends over time.

Results: In 2002, 51.8% (95% CI 46.0%, 57.3%) of children with CP were born LBW. By 2008, this percentage had declined, though not significantly, to 47.9% (95% CI 42.7%, 53.1%), p=0.51. In each surveillance year, the proportion of children with CP born LBW was higher for black non-Hispanic (BNH) than white non-Hispanic (WNH) children. For BNH children with CP the percentage born LBW decreased from 60.7% (95% CI 50.5%, 70.0%) in 2002 to 55.7% (95% CI 47.2%, 64.2%) in 2008, while for WNH children it decreased from 48.3% (95% CI 40.9%, 55.7%) in 2002 to 41.8% (95% CI 34.8%, 48.8%) in 2008. Between 2002 and 2008, the average annual percentage point changes among children with CP who were born LBW were -1.38% (95% CI -4.34%, +2.30%) for BNH, and -2.24% (95% CI -5.18%, +1.46%) for WNH children.

Conclusions: These data suggest that if the trends described here continued beyond 2008, the overall HP2020 objective for CP was likely met by 2013. In addition, absent a change in these trends, the racial disparity in the proportion of children with CP who were born LBW will not only persist but is likely to increase over time. Further examination of Hispanic and other ethnic groups is needed but could not be included here due to small numbers.

F8

Do persons with cerebral palsy from wealthy California zip codes live longer?

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Background/Objectives: In the general population, persons who live in areas with higher household incomes tend to live longer than those who live in lower incomes areas. We hypothesized that the same socio-economic gradient would be reflected in the cerebral palsy (CP) population.

Design: Observational cohort study.

who received services from the California Department of Developmental Services in years 1983 to 2010. We restricted the study to persons who lived in private households (rather than care facilities), and excluded those with degenerative disorders, autism, chromosomal anomalies, or traumatic injuries. Methods: We matched study participants to postal zip codes using administrative records. Functional skills for each individual were abstracted from the first Client Development Evaluation Report available for each patient during the study period. Participants were grouped by zip code-based median annual household income as measured in the 2000 United States Census. Descriptive statistics on age, sex and severity of disability were computed for each group. We estimated unadjusted survival probabilities with Kaplan-Meier survival curves, and estimated the effect of zip code, adjusted for age and functional skills, on mortality using Cox proportional hazards regression. Results: We identified 32 599 persons with cerebral palsy who lived in private households with known California zip codes. The proportions of individuals living in zip codes with median annual household incomes of <\$30,000, \$30,000 to 49,999. \$50 000 to 79 999, and >\$80 000 were 13%, 55%, 28%, and 4%. Age (mean 12.5, SD 12.3), sex (56% male), and the proportion who did not walk (GMFCS IV or V, 38%) were similar across zip codes. Collectively, participants contributed 459 199 person-years of follow-up, and 3336 died during the study period. Overall survival was 94% at 10 years and 87% at 20 years. Unadjusted survival was similar across the zip code-based income groups (log rank test, p=0.39). After adjustment for age, sex, walking skills, and the presence of a feeding tube, we still found similar mortality rates for the four income groups. The hazard ratios (HR) and 95% confidence intervals were: \$<30 000 HR 1 [reference], \$30 000 to 49 999 HR 1.0 [0.9-1.2], \$50 000 to 79 999 HR 1.0 [0.9-1.1], and > \$80 000 HR 1.0 [0.8-1.2].

Participants and setting: Persons with CP, aged 4 years or older,

Conclusions: Unlike in the general population, where persons who live in areas with higher income tend to live longer, the survival of persons with CP in California does not vary across zip codes with different levels of income. The results here, however, should be interpreted cautiously as we did not use direct measures of individual household income.

F9

Co-occurring autism spectrum disorder, intellectual disability, and epilepsy among children with cerebral palsy

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Background/Objectives: In 2006, the definition of cerebral palsy (CP) was changed to recognize that 'the motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication, and behavior, by epilepsy, and by secondary musculoskeletal problems'. While the co-occurrence of intellectual disability (ID) and epilepsy with CP is well-described, recent studies have shown that the prevalence of autism spectrum disorder (ASD) also is elevated in children

with CP. The purpose of this study was to assess the co-occurrence of ASD, ID, and epilepsy among children with CP.

Design: Cross-sectional, population-based surveillance program.

Participants and setting: Every 2 years from 2000 through 2010, children with CP were ascertained by the Metropolitan Atlanta Developmental Disabilities Surveillance Program (MADDSP), an active, multiple source, records-based surveillance system for ASD, CP, ID, hearing loss, and vision impairment. The study population includes 8-year-old children residing in the five county surveillance area during a given surveillance year. Trained clinical reviewers systematically code record abstractions and determine which children meet established case criteria for each disability. CP, ASD, and ID were ascertained for all surveillance years; co-occurring epilepsy was ascertained for children with CP beginning in 2006.

Methods: Chi-squared tests were used to assess differences in the frequency of ASD among children with CP by the presence of ID and epilepsy.

Results: From 2000 to 2010, 969 children with CP were ascertained, of whom 470 (48.5%) had co-occurring ID and 84 (8.7%) had co-occurring ASD. Among the 523 children with CP from 2006 to 2010, 232 (44.4%) had ID, 43 (8.2%) had ASD, and 219 (41.9%) had epilepsy. The remainder of the results reflect the 2006 to 2010 period. ASD frequency was 6.2% among children with spastic CP; 21.2% among children with non-spastic CP, and 10.3% among children with mixed/ other CP subtypes (p<0.01 for overall difference). Children with CP and ID were more likely to have ASD than those with CP without ID (11.2% vs 5.8%, p=0.03). In contrast, children with CP and epilepsy were less likely to have ASD than those with CP without epilepsy (5.0% vs 10.5%, p=0.03). The lower frequency of ASD among children with CP and epilepsy persisted when stratified by the presence of ID.

Conclusions: ASD occurs at a higher frequency among children with CP compared to the ASD frequency reported for the general U.S. population (1–2% in the most recent surveillance years). Universal screening for ASD and other developmental delays is important to ensure that all children, including those with CP, receive appropriate interventions. Further research is needed to understand whether the higher frequency of ASD among children with CP and ID and lower frequency of ASD among children with CP and epilepsy reflects etiology, ascertainment, or both.

F10

Patterns of gross motor impairment severity and motor type in preschool age children with cerebral palsy: comparison between high- and low-resource countries

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Background/Objectives: To compare the distribution of motor type and functional severity (GMFCS) in preschool-age

children with cerebral palsy (CP) in Bangladesh and Australia and examine associations with neonatal/postnatal complications and co-morbidities.

Design: Cross sectional, two cohort comparison.

Participants and setting: 301 children with CP aged between 18 and 36 months seen at tertiary or rehabilitation centres, 81 children born in Bangladesh (mean age=27.6mo, males n=50 (62%), and 220 children born in Queensland or Victoria, Australia (mean age=26.5mo, males n=141(64%).

Methods: Physicians (Australian trained) confirmed the diagnosis at entry (18–36mo). A detailed history was taken from the mother confirming gestational age (term, late preterm, very preterm and extremely preterm), timing of event (prenatal, perinatal or post natal) and neonatal/postnatal complications (apgar scores or delayed cry >5min after birth (Singhi et al, 2002) used to indicate birth asphyxia). Motor type (spastic, dyskinesia, hypotonia or ataxia by SCPE Guidelines) and Gross Motor Function Classification (GMFCS) by the same two independent raters via video analysis. Outcome and exposure variables between the two cohorts were compared using Chi-square and Chi-square Test for Trend. The association between motor outcomes (GMFCS and motor type) and birth history variables were explored using multinomial logistic regression with relative risk ratios (Stata 10.0).

Results: There was a significant difference in the distribution of GMFCS levels between the Bangladesh (GMFCS I-II=23.5%, III=30.9%, IV-V=45.7%) and Australian (GMFCS I–II=37.4%, III=13.6%, IV–V=29.1%) cohorts (χ^2 for trends=18.83 p<0.00). Similarly the distributions of motor type were significantly different for the Bangladesh (spasticity=61.7%, dyskinesia 27.2%, ataxia 11.1%) and Auschildren (spasticity=88.2%, dyskinesia=4.1%, ataxia=7.7%), with a higher proportion of children in the Bangladesh cohort diagnosed with dyskinesia and ataxia $(\chi^2=14.1, p<0.01)$. The Australian cohort had higher proportion of children born extremely/very pre term (26.8%) compared to the Bangladesh (3.9%), (χ^2 =25.9, p<0.01). Birth history complications including delay cry/low apgar scores <7 ($\chi^2=108.7$, p<0.01), jaundice ($\chi^2=31.2$, p<0.01), perinatal hospital admissions ($\chi^2=18.48$, p<0.01) and home deliveries $(\chi^2=96.4, p<0.01)$ were higher in Bangladesh. For the Australian cohort 5 minute apgar scores <7 were associated with motor severity (χ^2 =108.7, p<0.01), however delayed cry was not associated in Bangladesh. Home delivery was not associated with motor severity for the Bangladesh cohort, however in the Australian sample was associated with dyskinesia (RRR=12.0, p=0.05).

Conclusions: There is a striking difference in distribution of gross motor severity and motor type in preschool age children with CP between high and low resourced countries. This may be due to neonatal complications and has marked implications for the need and type of early rehabilitation.

F11

Change in level of lesions in individuals who have spina bifida in Southern Arizona in the preversus post-folate fortification eras

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Background/Objectives: Multiple randomized controlled studies have shown the efficacy of preconception folate supplementation for women in decreasing the incidence of spina bifida in the United States. Folate supplementation in grains was initiated in the United States in 1996 and was made mandatory in 1998. Anecdotally noted in a regional multispecialty referral center in southern Arizona was a change in level of lesion, with younger children having lower level of lesions compared with older children and adults. We reviewed clinic data over 27years to assess the validity of this observation.

Design: Retrospective medical record review.

Participants and Setting: Referral pediatric multispecialty center in southern Arizona.

Methods: The study is a retrospective cohort study including 192 medical records from clinic patients born between 1983 and 2010, excluding those patients born from 1994 to 1998. These patients were excluded because folate supplementation between 1994 and 1998 was optional. The study includes patients with a primary diagnosis of spina bifida, with or without hydrocephalus. Patients with spina bifida occulta were excluded from the review.

Results: Overall post-folate incidence of spina bifida was lower than the pre-folate period (22.4%). There were more children with high level of lesion (L2 and above) than expected in pre-folate and about 50% less than expected in post-folate periods (p<0.05). The proportion of wheelchair users is also significantly higher in the pre-folate group (p=0.05). The number of children who had mid-level lesions (L3–L5) and low-level lesions were distributed as predicted in pre- and post-folate supplementation. We found no differences between sex or race and pre- or post-folate periods, however Hispanic ethnicity classification was missing for a large proportion of the records (45.8%).

Conclusions: Nutritional folate supplementation has significantly decreased both the incidence and severity of spina bifida in a portion of southern Arizona through reduction in high level lesions. The younger children in this review have different clinical needs, with more children able to walk rather than use a wheelchair as their primary mode of ambulation.

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G1

Relationship between brain structure and communication skills in children with cerebral palsy

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Background/Objectives: To examine the relationship between the pattern and severity of the brain lesion on structural MRI (sMRI) and communication skills in preschool-aged children with cerebral palsy(CP).

Design: Prospective population based cohort study.

Participants and Setting: Of 139 children with CP (male n=79, 57%), mean age at clinical assessment was 28 months (5) and at sMRI 23 months (16). Gross Motor Functional Classification System(GMFCS) I=59(42%), II=17(12%), III=19(14%), IV=18(13%), V=26(19%).

Methods: Children were evaluated using the Communication and Symbolic Behavioral Scales Developmental Profile(CSBS-DP) Infant-Toddler Checklist and classified for GMFCS, motor type and distribution. sMRI scans were classified using (1) five qualitative Krägeloh-Mann(KM) patterns and (2) novel reliable semi-quantitative scale (total global score (GS)=40, Fiori) where higher scores indicate greater severity. Data was analysed using linear regression (SPSSv22).

Results: Total CSBS-DP standardized score for the cohort was mean (M)=86.8 (22.7), of these 66 children (48%) had delayed communication. Distribution of KM classification were: Periventricular White Matter(PWM)=89(64%), Cortical/Deep Grey Matter(CDGM)=26(19%), Brain Malformations(BM)=7 (5%), other miscellaneous lesions=10(7%), normal MRI=7 (5%). On univariate analysis the brain lesion severity (Fiori) GS was related to all aspects of communication (reported as β (95% CI): total CSBS-DP -1.0(-1.5, -0.6), social -0.2 (-0.3, 0.1), speech -0.2(-0.3, 0.0) and symbolic -0.2(-0.3, 0.0)-0.1) composites (all p<0.01). Overall severity of the brain lesion (GS) in relation to the domains of communication was more important than any of the hemispheric or lobar subscales (multivariate analysis). Brain lesion severity GS was related to gross motor function (GMFCS) (β =2.3 (1.5, 3.1) p<0.01). Children with poorer gross motor function also had poorer communication skills (F=16.0, p<0.01). In multivariate analysis brain lesion severity GS was more closely associated to gross motor function (β =1.9(1.0, 2.9), p<0.01) than to total communication ability (β =-0.04 (-0.1, 0.02), p=0.2). The pattern of the brain injury (KM) was related to expressive language skills (CSBS-DP speech composite) (β =-2.7 (-5.2, -0.2) p = 0.03). Children with PWM lesions had better speech composite scores M=9.1(5.6), than CDGM M=6.4(5.5), and BM M=5.13(2.6). There was no relationship between the pattern of brain lesion (KM) and the social and symbolic composites.

Conclusions: There is a relationship between both the brain lesion severity and pattern and communication skills in preschool-aged children with CP. In particular, the pattern of injury impacts on expressive language with children who have PWM lesions having better expressive language skills. As sMRI is routinely used in the diagnosis of children with CP, these results will help guide clinical decision-making and planning for early intervention services throughout the preschool years.

G2

Does early communication explain the relationship between motor ability and social function in children with cerebral palsy?

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Background/Objectives: To investigate whether early communication ability mediates the relationship between early motor ability and later social development in children with cerebral palsy (CP).

Design: Prospective population based cohort study.

Participants and Setting: Of 71 children with CP participated in the study (male *n*=43, 61%), Gross Motor Functional Classification System (GMFCS) distribution at 24 months corrected age (ca): I=24(34%), II=9(13%), III=12(17%), IV=10(14%), V=16(22%).

Methods: Children's motor ability was measured at 24 months ca on the Gross Motor Function Measure (GMFM) and GMFCS. Communication ability was evaluated at 24 months ca using the Communication and Symbolic Behavioral Scales Developmental Profile (CSBS-DP) Infant-Toddler Checklist which includes social, speech and symbolic composites. Social development was assessed at 60 months ca using the Pediatric Evaluation of Disability Inventory (PEDI). Data was analyzed using meditation bootstrap analysis (results are significant at 0.05 if 95% CI does not include 0) (SPSSv22).

Results: At 24 months ca 35(49%) children were identified with possible communication delay communication (>1SD below the mean on CSBS-DP). At 60 months 44(62%) children had social function scores on the PEDI >1 SD below the mean. Results on 10 000 bootstrapped samples indicated that the relationship between early motor ability (GMFM-66) and later social functioning (PEDI) was significantly mediated by early communication ability (95%CI=0.8, 0.42; b=0.24). Almost half (R²=0.47) the variability in social functioning was predicted by early motor function and communication ability. On analyses of the composites of the CSBS-DP, only the speech composite significantly mediated the relationship between early motor function and later social ability (95% CI=0.03, 0.34; b=1.0). The symbolic (95%CI=-0.09, 0.42;

b=0.82) and social composites (95%CI=-0.24, 0.18; b=-0.12) were not significant mediators.

Conclusions: These results demonstrate that early communication partially, but significantly mediates the relationship between early motor function and later social ability. Specifically the speech composite, that includes motor speech and expressive language, has an important role in this relationship. This finding highlights the significance of targeting early speech and language to support the development of social function. It emphasizes the need for further research into this relationship, particularly regarding the relative contributions of the parent-child communication dyad on social development and specific motor speech and language components of communication.

G3

Stability and validity of the Communication Function Classification System

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Background/Objectives: The Communication Function Classification System (CFCS) is an ordinal, five-level description of communication performance developed to provide caregivers and professionals an effective way to describe a child's every-day communication. Although, preliminary results are encouraging, further assessment of the validity and stability of this scale are needed. The objectives of this research were to: (1) Evaluate the stability of CFCS level over time (2) Evaluate the validity of the CFCS by comparing it to scores on standardized receptive/expressive language tests.

Design: Instrument Development & Validation.

Participants and Setting: Of 123 children with cerebral palsy who attend a yearly interdisciplinary cerebral palsy were included in this study. At the initial visit, the mean participant age was 6.4 years ([4.7] Range 1–20). Of the 123 participants, 86 children had a standardized language test.

Methods: A speech-language pathologist assigned a CFCS level at every visit and administered an age appropriate standardized language test at most visits. Standardized tests included the Preschool Language Scale (PLS-5); and the caregiver interview Receptive-Expressive Language Test (REEL-3). Spearman correlation coefficients were used to compare CFCS stability over time.

Results: (1) 76% (94/123) of the children were classified with the same CFCS level at subsequent visits (r_s =0.93; p<0.0001 for visits 1–2 and r_s = 0.95; p<0.0001 for the 66 children with 2–3 visits). 7 children decreased a level (i.e., demonstrated less functional communication); 17 children improved 1 level and 5 improved 2 levels (i.e. demonstrated more functional communication). There was no difference in age between the group of participants who did not change levels (N=94; mean age=6.6y [4.3]) and those that did change (N = 29; mean age=6.1y [4.9]). (2) Standardized language assessments on 86 children were evaluated by CFCS level. The mean PLS total standard scores declined in each less

functional CFCS level. The standard scores were significantly different for CFCS levels II and III (k=3; p=0.026) (mean=87.4 [16.3]; mean=73.4 [11.2] respectively) but not between CFCS levels I and II (mean level I=92.8 [14.8]) (k=1; p=0.31) or III and IV (mean level IV=66.7 [14.3]) (k=2; p=0.31). The PLS was not routinely administered to children assigned a CFCS level V.

Conclusions: The CFCS level remained stable for 76% of children with CP. Age or initial CFCS level was not significantly different between the groups of children who did and did not change CFCS levels. Standardized language tests appeared to differentiate between levels. This is the first study to compare the CFCS level over time and suggests that the CFCS may be stable for a significant number of children with cerebral palsy. Additional research is needed to increase the number of children studied and the length of time.

G4

Predicting communication functioning at school entry in children with cerebral palsy

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Background/Objectives: To examine demographic, environmental and early comorbid variables as predictors of communication functioning at school entry in children with Cerebral Palsy (CP).

Design: Prospective cohort study.

Participants and Setting: Of 114 children with CP (76 male, 66%) who received early (18 or 24mo) and late assessments (48 or 60mo) were recruited in a population based sample. Gross Motor Functional Classification System (GMFCS) at 18/24 months was I=42 (37%); II=14(12%); III=17(15%); IV=20(18%); V=21(18%) and 48/60 months: I=50(44%); II=14(12%); III=12(11%); IV=23(20%); V=15(13%). Communication functioning at 48/60 months on the Communication Function Classification System (CFCS) was: I=36(32%); II=25 (22%); III=20(18%); IV=19(17%); V=14(12%).

Methods: Early communication was evaluated using the Communication and Symbolic Behavioral Scales Developmental Profile (CSBS-DP) Infant-Toddler Checklist and classified by an allied health professional at 48/60 months using the CFCS. A Physiotherapist classified GMFCS, Manual Ability Classification System (MACS), motor type, and distribution at 18/24 and 48/60 months. Demographic (sex, socioeconomic status, SES) and comorbid variables (birth-order, multiple gestation, vision, hearing, epilepsy, prematurity, Alternative and Augmentative Communication (AAC) were obtained through parent interview by a medical specialist. A multivariate multinomial regression was used to analyze the value of early communication, GMFCS, MACS and demographic/comorbid variables in predicting communication on the CFCS at school entry (SPSSv22).

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Results: At 18/24 months the CSBS-DP standard score was mean 84.9 (19) with 50 children (43.9%) identified with communication delay. Nine (8%) children used some form of AAC. At 18 to 24 months only CSBS-DP (χ^2 =42.7, p<0.01) and GMFCS (χ^2 =27.2, p<0.01) were strong predictors of communication functioning at school entry, not demographic variables or comorbidities (multivariate analysis). Poorer scores on CSBS-DP at 18/24 months were associated with lower functional classification on CFCS at 48/60 months (CFCS I is the reference, reported as Odds Ratios (95% CI): CFCS II OR=0.8(0.5-1.1) p=0.15; III OR=0.4(0.2-0.7) p<0.01; IV OR=0.1(0.02-0.3) p<0.01; V=NS). Lower motor function classification on GMFCS was associated with poorer communication (CFCS II OR=1.4(0.8-2.5) p=0.23; III OR=2.9(1.5-5.6) p<0.01; IV OR=4.9(2.0-12.0) p<0.01; V OR=14.7(3.5-62.3) *p*<0.01).

Conclusions: Unlike their typically developing peers, in children with CP biological factors, particularly gross motor attainment and early communication skills, have a greater impact on communication ability at school entry rather than environmental influences, such as SES and birth-order. This has important implications for targeting early interventions/ support for communication ability at school entry.

G5

Rehabilitation services for preschool children with primary language impairment: individual versus dvad intervention

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Background/Objectives: Children with primary language impairment (PLI) require rehabilitation services to facilitate development and community participation. Long waiting times for services suggest that new approaches to service provision are necessary, while maintaining treatment effectiveness. The primary objective of this study was to compare the effectiveness of two rehabilitation service delivery approaches (dyad versus individual) for young children with moderate to severe PLI on overall development, adaptive behavior, social skills at school and parent satisfaction.

Design: Randomized controlled trial.

Participants and Setting: Children 3 to 4 years of age referred to a rehabilitation hospital pediatric speech-language program with a diagnosis of moderate to severe PLI were eligible. Children with cognitive deficits, autistic features or hearing impairment were excluded.

Methods: Participants were randomized to receive either dyad (2 children to 1 therapist) or individual (1 child to 1 therapist) intervention over 1 year, using a stratified (age, severity) block design. Dyad groups were created according to type of language impairment, language of intervention, child's age and

overall functioning, and intervention in both groups was provided as usual. Outcome measures were conducted blindly and included assessments of development (Battelle Developmental Inventory [BDI]), adaptive behavior (Vineland Adaptive Behavior Scales [VABS]), expressive language (Mean Length of Utterance [MLU]), and receptive language (Peabody Picture Vocabulary Test [PPVT-III]), parent satisfaction (Measure of Processes of Care [MPOC]) and social performance at school or day care (Social Skills Rating System [SSRS]).

Results: Overall, there were no significant group differences between dyad and individual interventions on measures of development, speech, social skills and parent satisfaction. Only mean individual change in the motor domain (BDI) over the year of intervention was significantly greater for those in the dyad group (dyad: +6.16; individual: -1.38; *p*=0.05) while change in PPVT scores was significantly greater for children who received individual treatment (dyad: +3.7; individual: +14.6; *p*=0.0003).

Conclusions: Similar outcomes between children who received dyad and individual intervention indicate no detrimental effect of the dyad approach and suggest this may be used as an alternative to traditional individual intervention, potentially as a cost-effective strategy to decrease wait times.

G6

Jaw kinematics of chewing in children with cerebral palsy

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Background/Objectives: Children with CP have more frequent choking/coughing during mealtimes and are introduced to solid foods at later ages as compared to their typically-developing (TD) peers. A reason for the feeding difficulties may be immature jaw motion. Quantitative accounts of chewing development in TD children suggest that immature mastication is characterized by inefficient jaw movements, such as increased speed, path distance, and duration. We hypothesize that children with CP will have increased speed, duration, path distance, and volume when compared their TD peers. Design: Cross-sectional design.

Participants and Setting: A volunteer sample of 15 children with spastic CP (GMFCS levels I to V; 10 M, 5 F) and 15 age-and sex-matched typically-developing (TD) peers between the ages of 2;8 and 18;6 years of age were recruited for this study. Participants in the TD group had no history of speech, language, cognitive, neurological, or motor impairments.

Methods: Participants were recorded with an 8-camera optical motion capture system. Three small spherical reflective markers were placed on the chin. A rigid plate with 4 markers was attached to the forehead to subtract head movement from the jaw data. Chewing sequences were parsed to include only the mid 80% of each sequence to eliminate extraneous jaw motion. Participants ate at least 5 trials of 3 consistencies: solid (Cheerios), mechanical soft (banana), and puree (applesauce). Bolus size was maintained across trials and participants to 1 teaspoon. The duration (s), average speed (mm/s), and

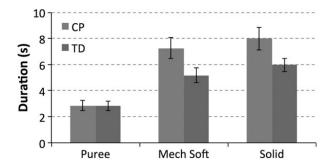


Figure 1: Duration in seconds of puree, mechanical soft, and solid consistences of children with cerebral palsy and their typically-developing peers.

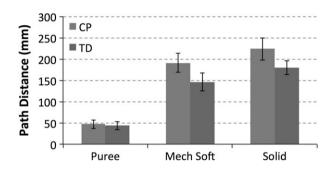


Figure 2: Path distance in millimeters of puree, mechanical soft, and solid consistences of children with cerebral palsy and their typically-developing peers.

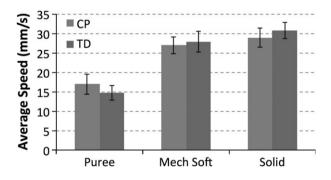


Figure 3: Average speed in mm/s of puree, mechanical soft, and solid consistences of children with cerebral palsy and their typically-developing peers.

path distance (mm) of each trial was obtained using custom Matlab algorithms. A 2-standard-deviation ellipse was fit around the path distance to quantify the volume (mm³) of jaw motion for each sequence. A 2×3 repeated-measures ANCO-VA was conducted to examine the effect of Group (CP, TD) and Consistency (solid, mechanical soft, puree) with age and sex as a covariate on each of the kinematic variables.

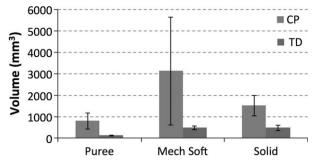


Figure 4: Volume in mm3 of puree, mechanical soft, and solid consistences of children with cerebral palsy and their typically-developing peers.

Results: Children with CP had greater durations for mechanical soft (CP: mean (M)=7.25s, standard error (SE)=0.80s; TD: M=5.18s, SE=0.56s, p<0.001) and solids (CP: M=7.99s SE=0.87s; TD: M=5.97s, SE=0.51s p<0.001). The CP group also had greater path distances for mechanical soft (CP: M=190.58mm, SE=22.23mm; TD: M=146.15mm, SE=21.01mm, p<0.001) and solids (CP: M=223.56mm, SE=26.42mm; TD: M=179.09mm, SE=16.37mm, p<0.001). The CP group had greater movement volumes for puree (CP: M=791.73mm³, SE=377.03mm³; TD: M=112.54mm³, SE=28.14mm³, p<0.001), mechanical soft (CP: M=3114.94mm³, SE=2507.72 mm³; TD: M=329.88mm³, SE=85.17mm³, p<0.01) and solids (CP: 1518.39mm³, SE=483.56mm³; TD: M=471.02mm³, SE=125.65mm³, p<0.001). No group differences were observed for average speed.

Conclusions: Children with CP had longer durations, greater path distances, greater volumes than the TD peers which may reflect difficulties with oromotor control during chewing and may have implications for other oromotor tasks (e.g., speech).

G7 Oropharyngeal dysphagia in preschool children with cerebral palsy: comparison between highand low-resource countries

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Background/Objectives: Feeding difficulties (oropharyngeal dysphagia, OPD) are common in young children with cerebral palsy (CP) based on studies in high-resource countries. There has been no analysis of how patterns of OPD differ between high- and low-resource countries.

Objective: To determine the prevalence and severity of OPD in preschool children with CP in Bangladesh, and how this compares to Australian children.

Design: Cross sectional, comparison of two cohorts.

Participants and Setting: Of 211 children with CP aged between 18 to 36 months, 81 children born in Bangladesh

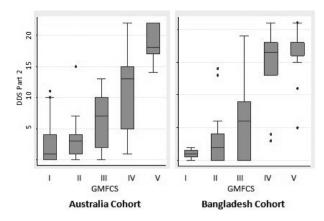


Figure 1: Dysphagia Disorders Survey scores by GMFCS level for Australia and Bangladesh Cohorts.

(mean age=27.6 months, 61.7% males), and 130 children born in Queensland, Australia (mean age=27.4 months, 62.3% males). Cohorts were stratified according to ambulatory status for analysis (GMFCS I-II, III, IV-V).

Methods: OPD prevalence and severity were analyzed using Part 2 of the Dysphagia Disorders Survey (DDS) (scored out of 22) rated from video by a speech pathologist. Gross motor skills were classified using the Gross Motor Function Classification System (GMFCS), motor type and distribution. Data were analyzed using (1) descriptive statistics (GMFCS, motor type, distribution; prevalence (%) of OPD (adjusted based on validation data); mean DDS score, SE); tube feeding (2) Difference in OPD prevalence between Bangladesh and Australia samples by GMFCS strata using chi2 and binomial logistic regression; (3) Difference in OPD severity between Bangladesh and Australia cohorts by GMFCS strata using independent t tests and linear regression (Stata v10.0).

Results: (1) Bangladesh (BD) sample (%): GMFCS I to II=23.5, III=30.9, IV to V=45.7, Motor type: spasticity=61.3, dyskinetic=27.5, ataxia/hypotonia=11.3; lence=66.6%; OPD severity=10.4 (SE=0.9); no tube feeding. Australia (AU) sample (%): GMFCS I to II=56.2, III=17.7, IV to V=26.2, Motor type: spasticity=86.8, dyskinetic=4.7, ataxia/ hypotonia=7.8; OPD prevalence=33.3%; OPD severity=7.0 (SE=0.7); tube feeding=10.8%. (2) There was no difference in OPD prevalence between cohorts stratified for motor severity: GMFCS I-II-AU=31.5%, BD=31.6% (p=0.99); GMFCS III-AU=69.6%, BD=56.0% (p=0.33); GMFCS IV-V-AU=91.2%, BD=91.9% (p=0.91); OPD prevalence was associated with GMFCS (OR=4.6, p<0.01). (3) There was no difference in OPD severity (DDS score) between cohorts stratified for motor severity (Fig 1): GMFCS I to II-AU=2.96, BD=2.88 (p=0.94); GMFCS III-AU=6.3, BD=5.7 (p=0.73); GMFCS IV to V-AU=16.4, BD=16.0 (p=0.71); OPD severity associated with GMFCS (F=86.8, p<0.01).

Conclusions: Despite qualitative differences in the patterns of OPD between the Bangladesh and Australian samples, the prevalence (when adjusted for the functional gross motor severity of the cohorts) was equivalent. Furthermore, the severity (based on the sum of the number of impaired ingestion functions) was also comparable between cohorts (when

adjusted for GMFCS). This study provides support for the robust association between functional motor severity and OPD prevalence/ severity in young children with CP, regardless of ethnicity and health resourcing.

G8

Development of work participation in young adults with cerebral palsy: a prospective study

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Background/Objectives: Youth with cerebral palsy (CP) nowadays makes the transition to adulthood; little is known about their development of work participation over time. This study aimed to assess the development of work participation among young adults with CP transitioning into adulthood, and to examine associated characteristics and perceived work limitations and barriers.

Design: Prospective cohort study over a 4-year period.

Participants and Setting: Participants were young adults with CP of average intelligence (n=74), known at eight rehabilitation centers in the Netherlands, aged 16 to 20 years at baseline. Mean age at 4-year follow-up was 22.8 years (SD 1.4y), 61% was male, and 83% was classified at GMFCS level I to II.

Methods: We performed structured interviews at baseline, 2-year and 4-year follow-up. We assessed work participation in three categories (employed, unemployed, studying). At 4-year follow-up we examined associations of this outcome with demographic and clinical characteristics using multinomial logistic regression. At this time we inventoried work limitations and barriers among employed persons, using the Work Limitations Questionnaire and a 28-items checklist of barriers.

Results: From age 16 to 20 years to age 20 to 24 years the proportions employed and unemployed persons increased from 12 to 49% and 3 to 17% respectively; the proportion students decreased from 85 to 34%. At the age of 20 to 24 years, those with a lower age and with a lower GMFCS level were more likely unemployed than employed (adjusted Odds Ratio 0.30 and 0.08 respectively). The employment rate of young adults with CP aged 20 to 24 years was lower than in the general population of the same age (59%). Employed persons experienced few work limitations; 28% perceived situational or health barriers.

Conclusions: Young adults with CP and average intelligence seem to be at risk for an unfavorable development of work participation. Rehabilitation services might offer support to prevent work disability and unemployment.

G9

Motor imagery and planning deficits in children with congenital hemiplegia

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Background/Objectives: Motor imagery (MI), the mental simulation of an upcoming movement, involves neural networks that partially overlap those involved in motor planning and control (MC). MI training has shown promising results in adult stroke patients, who often retain accurate MI abilities; however, emerging evidence suggests that MI may be compromised in children with congenital hemiplegia. The purpose of this study was to use objective tests to evaluate MI functions in children with congenital spastic hemiplegia and how these relate to standard evaluations of upper extremity motor performance.

Design: Prospective cohort.

Participants and Setting: Of 18 participants with congenital hemiplegia (HP) (MACS I-II, M=15y), 18 matched peers (PR) were recruited from a Children's hospital and the community. Methods: Motor imagery speed and accuracy were assessed with the computer based Prospective Grip Selection (PGS) task.1 Motor performance was evaluated with the fine motor subtests of the Bruininks-Oseretsky Test of Motor Proficiency (BOT-2). For the MI and MC conditions, participants were presented with 3D disk, with two opposing contact points, presented in 12 orientations (30° steps around a full circle). In the MI condition participants verbally indicated their grip preferences while remaining still. In the secondary MC condition participants chose either an over- or under-hand pincer grip to grasp the widget on the contact points. In MC and MI, both hands were tested in counterbalanced order across participants. MI mean response times (RTs) were computed separately for each hand and each stimulus orientation (0-330°). MI accuracy was evaluated by quantifying similarity with preferences exhibited in MC. The subtests of Fine Motor Precision (FMP), Fine Motor Integration (FMI) and Manual Dexterity (MD) were performed per protocol. Data were analyzed using ANOVAs, t-tests, and Pearson's correlations. Results: MI RTs were significantly slower for the HP group (F (1,66)=14.4, p<0.001) with angle of orientation producing significant variability in the HP group only (F(11,24)=3.1,p=0.01) (Chart 1). Between hand differences were not significant in either group. Accuracy was significantly different across angles of orientation (F(11,58)=7.1, p<0.001); however, between group and hand differences were not significant (Chart 2). The HP group scored significantly lower than peers on all BOT-2 subtests (Table 1). The MI task RTs correlated significantly with FMP and MD subtests for the HP group, and the FMI subtest for the PR group (Table 2).

Conclusions: The PGS task suggests that children with congenital hemiplegia retain the ability to imagine grasping movements in a manner that accurately reflects their current motor capabilities, albeit more slowly than peers. MI accuracy is furthermore related to difficulties in functional motor perfor-

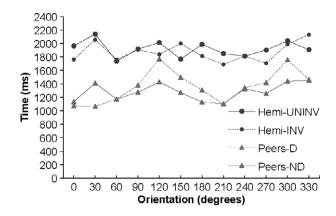


Figure 1: Chart 1: Motor Imagery Reaction Time

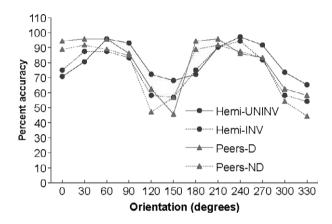


Figure 2: Chart 2: Agreement between Motor Imagery and Motor Control

mance. These findings raise the possibility of employing MI as a complimentary therapy in this population. ¹Johnson, S. Thinking ahead: the case for motor imagery in prospective judgments of prehension. *Cognition* 2000; **74**; 33-70.

Table 1: BOT-2 Fine Motor Subtests (means, SD)

	Hemiplegia	Peers	
Fine Motor Precision	38.8 (2.2)	40.6 (.8)	p=.003
Fine Motor Integration	36.3 (3.1)	38.6 (1.5)	p=.04
Manual Dexterity	25.2 (5.5)	34.8 (4.1)	p<.001

Table 2: BOT-2 Fine Motor Subtest and Motor Imagery correlations

	Hemi	plegia	Pe	ers
		Motor i	magery	
	NonInvolved	Involved	Dominant	Nondominant
Fine Motor Precision	r =6	r =4	ns	ns
	(p=.01)	(p=.05)		
Fine Motor Integration	ns	ns	r=7	r =6
			(p=.002)	(p=.02)
Manual Dexterity	r=7 r=5		ns	ns
•	(p=.01)	(p=.03)		

G10

Early versus later brain injury: no difference in executive functioning

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Background/Objectives: Objective: It has been argued that early brain insult results in less significant neuropsychological impairment because the developing brain is capable of greater plasticity and reorganization (Anderson et al., 2005). The current study aims to evaluate this claim by comparing executive functioning (EF) following early (Congenital Hemiplegia) versus later Acquired Brain Injury (ABI).

Design: Study Design: Comparison between two cross-sectional cohorts and a typically developing control group.

Participants and Setting: Method: 95 children with unilateral cerebral palsy (UCP); Male=43; mean (±SD) age=11.81 (2.3) years; range 8 to 17 years; GMFCS I=46, II=49; MACS I=25, II=70, IQ=82.06 (17.93); 56 children with ABI (Male=32; age=11.69 (2.4) years; equivalent to GMFCS I=26, II=30; equivalent to MACS I=20, II=36, IQ=76.33 (15.94) and 19 typically developing children (TDC); 9 male; mean age (SD) =10.8 (2.4) years; range 8 to 16 years; IQ=116.11 (10.8) were included.

Methods: Four domains of EF: attentional control (WISC-IV Digit Span Backwards); cognitive flexibility (DKEFS Colourword Inference Task); goal setting (Tower of London Dx and DKEFS Tower); and information processing (WISC-IV Symbol Search). Between-group differences (CP vs. ABI) were analysed using ANCOVA with age as a covariate (SPSS 21). Results: The ANCOVA for each domain of EF between groups was significant: attentional control F(3,166)=12.44, p>0.001; cognitive flexibility F(3,166)=4.81, p=0.009; goal setting F(2,164)=9.51, p>0.001; and information processing F (3,165)=9.73, p>0.001. Follow-up pairwise comparisons revealed a significant difference between TDC and CP across each domain including: attentional control (mean difference, MD=-1.85, p>0.001), cognitive flexibility (MD=18.58), p=0.01), goal setting (MD=-3.06, p>0.001) and information processing (MD=-7.78, p>0.001) as well as a significant difference between TDC and ABI groups, across each domain including; attentional control (MD=-2.19, p>0.001), cognitive flexibility (MD=24.29, p=0.002), goal setting (MD=-2.47, p=0.001) and information processing (MD=9.82, p>0.001). However, no significant difference was observed between ABI and CP groups on any domain: attentional control (MD=0.34, p=0.31), cognitive flexibility (MD=-5.64, p=0.26), goal setting (MD=0.59, p=0.22) and information processing (MD=2.03,

Conclusions: Conclusion: Regardless of age of injury, both children with congenital hemiplegia and those with later ABI demonstrated diffuse impairment across all domains of executive functioning. These findings have profound clinical implications as currently children with UCP have limited access to neuropsychological intervention. Yet, this study suggests that

they demonstrate very similar impairments to children with ABI, for whom neuropsychological evaluation is standard care. Such compromised cognitive functioning has the potential to impact on current and future education, employment and social integration.

Free Papers H

H1

Wired for recovery? How corticospinal system connectivity impacts on the efficacy of bimanual therapy in children with unilateral cerebral palsy

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Background/Objectives: To examine whether corticospinal tract (CST) connectivity has an influence on the efficacy of intensive bimanual therapy in children with unilateral spastic cerebral palsy (USCP). Reorganization of motor pathways can occur after early brain injury. In some children, ipsilateral CST projections from the contralesional hemisphere control the impaired arm, while in other children the typical contralateral CST projection pattern is established. Previous studies have shown that CST connectivity pattern influences the efficacy of constraint-induced movement therapy (CIMT). We here compare the efficacy of intensive bimanual therapy between children with ipsilateral vs. contralateral CST projections controlling the impaired hand. Hypothesis: the efficacy of bimanual therapy is different for children with contralateral vs. ipsilateral CST projections.

Design: Prospective cohort-study.

Participants and Setting: Thirty-one children with USCP, university laboratory day-camp.

Methods: Hand-Arm Bimanual Intensive Therapy (HABIT) was performed in a day-camp setting for 3 weeks, 5 days a week, 6 hours a day (total of 90h). Children were involved in a variety of bimanual play and functional activities in which they had to actively use both hands. To assess the effects of HABIT, hand function was tested before and after the intervention with the Jebsen-Taylor Test of Hand Function (JTTHF), a timed measure of unimanual dexterity. We used single-pulse transcranial magnetic stimulation (TMS; Magstim 200, figure 8-coil) to identify the cortical representation of the impaired arm and hand. Muscle activity was measured bilaterally with electromyography (EMG) from the first dorsal interosseus, wrist flexors, and biceps muscles. TMS stimulation sites were defined as responsive when they elicited a motor evoked potential (MEP) $>50\mu V$ in one or more of the three muscles. When stimulation of the affected hemisphere elicited responses in the impaired arm, the participant was classified as having a contralateral CST. When responses in the impaired arm were only found when stimulating the non-affected hemisphere, the participant was classified as having an ipsilateral CST.

Results: There were no differences in baseline JTTHF between children with a contralateral versus an ipsilateral CST controlling the affected hand (p=0.44). Overall there was a significant improvement in movement speed after 90 hours of HABIT (p<0.001). However, children with an ipsilateral CST (n=18) showed a trend towards greater improvement on the JTTHF than children with a contralateral CST (n=13; p=0.055).

Conclusions: The efficacy of HABIT seems to be related to the CST connectivity in children with unilateral CP. This finding is important for the optimization and tailoring of intervention programs for children with USCP.

H2 Hand function in a total age cohort of children with cerebral palsy in Norway

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Background/Objectives: Children with cerebral palsy (CP) in Norway are systematically followed up with both specialized and general health care. The CP-follow-up program (CPOP) was implemented in Norway in 2006, and register hand function and interventions aiming at the prevention of functional limitations in upper extremities. The aim of this study is to show the change in hand function between two registrations in the follow-up program and associations with given interventions.

Design: A population based longitudinal study.

Participants and Setting: A total population of children with CP born in 2006 in Norway. Their first assessment was at 4 to 6 years of age (mean 4.6) and the second was 1 to 3 years later (mean age 6.4).

Methods: The children are assessed at their habilitation units once or twice a year, or every second year, depending on age and functional level. The standardised CPOP protocol includes CP subtypes, Manual Ability Classification System (MACS), House functional classification, range of motion, spasticity, and interventions towards hand function, BoNT-A injections, orthoses, hand surgery and hand therapy such as treatment in bimanual activities, constraint-induced therapy or supervision and adaptations for bimanual use.

Results: Totally 128 children are included, representing a prevalence of 2.2. per 1000 live births. Classification according to SCPE shows; bilateral CP 38%, unilateral CP 47%, dyskinetic CP 11%, ataxic CP 4%, and 1% are not classified. MACS classification registered after 4 years of age shows the following distribution: Level I 37%, level II 27%, level III 9%, level IV 9% and level V 19%. House functional classification (scale 0-8) shows that the most affected hand is spontaneously used in 42% of the children (level 7-8), is an active assisting hand in 30% (level 5-6), a passive assisting hand in 21% (level 1-3) and is not used in 7% (level 0). After 1 to 3 years twenty one children (18%) have improved and ten children (8%) have a decrease in hand function according to House. There is a significant correlation between change in hand function and MACS level (p=0.001) Interventions: 53% of the children have hand therapy, 27% of the children use orthoses, 15% are treated with BoNT-A injections, only 1 child has undergone hand

surgery. MACS I-III; Significant correlation between improvement in hand function according to House and hand therapy is significant (*p*=0.003), and orthoses (*p*=0.008) and BoNT-A (*p*=0.018). MACS IV-V; No correlation between change in hand function according to House and given interventions. *Conclusions:* The change of hand function according to House functional classification in a total age cohort of children with CP in Norway after 1 to 3 years shows a significant improvement on MACS level I to III. There is a significant correlation between the improved hand function and given interventions.

H3 Dexterity of the unaffected hand in children with hemiplegic cerebral palsy

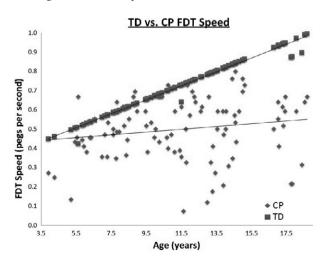
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Background/Objectives: There is a large amount of research on the rehabilitation of the affected upper limb in children with spastic hemiplegic cerebral palsy (CP). Two current methods, constraint-induced therapy and bimanual therapy, were derived from studies in adults with unilateral deficits due to cerebrovascular events and with an unaffected contralateral upper limb. However, data regarding the function of the unaffected upper limb in children with unilateral CP is lacking. The purpose of this study was to answer two questions: (1) Is the unaffected hand truly unaffected; that is, does it demonstrate dexterity similar to that of typically-developing (TD) peers? (2) What is the rate of dexterity acquisition in the unaffected hand in children with hemiplegic CP?

Design: Cross sectional study.

Participants and Setting: Consecutive series of 66 children (mean age 11.5y, range 3–18 y; 39 boys, 27 girls) with spastic hemiplegic CP followed in a pediatric CP specialty clinic. Methods: The Functional Dexterity Test (FDT) was used to measure the dexterity of the unaffected hand in all study participants. FDT speed was compared to published pediatric norms. Subgroups based on MACS, birth weight and gestational age were also compared.



Results: FDT speed of the unaffected hands is similar to normative values at 4-to-6 years of age (0.383 pegs/sec vs. 0.485 pegs/ sec, p=0.07) and this value increases linearly with age. However, the rate (dexterity acquisition) is 0.025 pegs/ sec/year slower than that of typically-developing (TD) children (0.010 pegs/sec/year vs 0.035 pegs/sec/year), resulting in a large difference by 17-to-18 years of age (0.511 pegs/sec vs 0.937 pegs/sec, p < 0.05). Within our subgroup analysis, we found that both birth weight and prematurity affected FDT speed. While all CP children had slower FDT speeds than TD norms, CP children who were born at <37 weeks gestation had a greater mean difference from age-matched norms than did children born at 37 weeks or later (0.255 pegs/sec vs 0.181 pegs/sec, p < 0.05). Also, CP children with higher birth weights (from 0.25-kg to 4.56-kg) tended to have higher FDT speeds (p<0.05 in each weight subgroup).

Conclusions: Our data shows that as children with spastic hemiplegic CP grow and develop manual dexterity, their unaffected hands fail to reach the level of their TD peers, despite starting from a similar point. Their achieved dexterity is related to age, birth weight, and prematurity. This data demonstrates the importance of assessing and including the 'unaffected' hand specifically when planning rehabilitation strategies, and perhaps limiting the amount of forced constraint or disuse.

H4

Poor description of upper limb therapies for children with unilateral cerebral palsy: a barrier to uptake of evidence into practice

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Background/Objectives: Incomplete description of interventions in randomized trials limits the uptake of therapies with proven efficacy into clinical practice. Poor reporting of comparison interventions hampers interpretation of treatment effect magnitude. This study aimed to evaluate the completeness of reporting of research interventions and control/comparator interventions in trials of upper limb therapies for children with unilateral cerebral palsy.

Design: Secondary analysis of trials in a systematic review¹ and subsequent publications of non-surgical upper limb therapies for children with unilateral cerebral palsy.

Participants and Setting: Randomized controlled trials of upper limb therapies in cerebral palsy.

Methods: Forty-seven randomized trials and 9 protocol papers were included, encompassing 47 research interventions and 55 control interventions. Using the 12-item Template for Intervention Description and Replication (TIDieR) checklist², 2 reviewers independently rated intervention and control descriptions by assessing the primary trial report, published protocols, and any online supporting materials and websites. Checklist items include: rationale, materials, procedures, intervention provider, mode of delivery, location of intervention, when and how much of the intervention (i.e. duration/dose, scheduling), tailoring, and treatment fidelity.

Results: The procedure component of interventions was adequately reported for 64% of research interventions and 31% of control interventions. Materials used in the research interventions (e.g. home program logs, splints) were mentioned in 80% of trials, yet only 13% of trials provided sufficient details to access or replicate the materials. Training materials used with intervention providers were mentioned in 32% of trials, yet only 4% reported access details to the materials. Sufficient detail about the location of where the intervention was provided was included for 55% of research interventions and 36% of control interventions. More than one clinician provided the research intervention in 53% of trials, yet intervention fidelity was assessed in only 40% of trials.

Conclusions: Few research interventions were described with sufficient detail to enable replication of the intervention, with crucial details missing in many. Even poorer reporting occurred for control interventions. Lack of comprehensive reporting of interventions contributes to worldwide waste in research funding, limits uptake of research findings in clinical practice, and hampers evidence synthesis. Authors, reviewers, and editors need to improve the quality of intervention reporting in published trials. The TIDieR checklist and guide is a potential solution and makes it easier to structure accounts of interventions. 1Sakzewski L et al. Efficacy of upper limb therapies for unilateral cerebral palsy: A metaanalysis. Pediatrics 2014, 133:e175-204. ²Hoffmann T et al. Better reporting of interventions: the Template for Intervention Description and Replication (TIDieR) checklist and guide. BMJ (in press, 2014).

H5

Hand and Arm Bimanual Intensive Therapy Including Lower Extremity (HABIT-ILE) in children with cerebral palsy: a randomized trial

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Background/Objectives: Hand-arm bimanual intensive training (HABIT) has shown evidence of efficacy for improvement in upper extremity (UE) use in children with unilateral spastic cerebral palsy (USCP). However, it is not known whether adding a lower extremity component to bimanual training is beneficial. Objective: To examine the efficacy of Hand and Arm Bimanual Intensive Therapy Including Lower Extremity (HABIT-ILE) for children with USCP.

Design: A randomized trial with a cross-over between HABIT-ILE and a control group.

Participants and Setting: Twenty four children with USCP were randomized, with stratification by age, sex, hemiparetic side and dexterity, to one of the two study arms. In phase 1, 12 children were assigned to the immediate HABIT-ILE group (IHG, 10d, for a total of 90h), and 12 to the delayed HABIT-ILE group (DHG), which continued to receive their traditional/ongoing treatment for an intended amount of 90 hours. Both

groups were assessed before and after their respective treatments. In phase 2, children in the DHG were crossed over to receive HABIT-ILE and children of the IHG were followed for an equivalent amount of hours of conventional therapy.

Methods: Manual ability was assessed using the Assisting-Hand-Assessment (AHA-primary outcome), the ABILHAND-Kids and the Pediatric Evaluation of Disability Inventory. Dexterity (Box and Blocks test) and pinch strength were also measured. Locomotor abilities were assessed with the ABILO-CO-kids and the 6 minute Walk Test (6MWT-primary outcome). Social participation was measured with the Life-HABITs (Life-H).

Results: Using a 2 (groups) \times 3(test session) ANOVA_{RM}, primary outcomes (AHA, p<0.001; 6MWT, p=0.002) and most secondary assessments (pinch strength, ABILHAND-kids, ABILOCO-Kids and social participation with Life-H) presented significant improvements following HABIT-ILE (all p<0.05), but not conventional therapy.

Conclusions: This first attempt to add a systematic lower extremity component to intensive bimanual training suggests that this combined approach can be useful for improving both upper and lower extremity function in children with USCP.

H6

Repetitive transcranial magnetic stimulation for perinatal stroke-induced cerebral palsy: short-term outcomes from the PLASTIC CHAMPS trial

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Background/Objectives: Perinatal stroke causes hemiparetic cerebral palsy. Developmental plasticity models have informed central therapeutic targets. Repetitive transcranial magnetic stimulation (rTMS) can modulate motor learning in adult stroke but is untested in perinatal brain injury.

Design: PLASTIC CHAMPS (www.clinicaltrials.gov/NCT01189058) was a randomized, blinded factorial trial of rTMS and constraint therapy (CIMT) in stroke-induced cerebral palsy.

Participants and Setting: Children 6 to 18 years participating in a 2 week goal-directed motor learning camp were randomized to inhibitory rTMS (1Hz, 1200 stimulations) over non-lesioned motor cortex, CIMT, both or neither.

Methods: Primary short-term outcome was 1 week post-camp Melbourne Assessment (MA). Additional outcomes included Assisting Hand Assessment (AHA), Canadian Occupational Performance Measure (COPM), and safety/tolerability. Corticospinal tract (CST) orientation was defined using singlepulse TMS.

Results: All forty-five children completed the trial (median 11.4y, mean MA(SD) 74(23)%). All improved on multiple measures at 1 week. Compared to sham, rTMS was associated with greater gains in MA (5.51(5.9) vs 1.89(4.7), p=0.027). CIMT did not affect 1 week MA. Group comparison suggested greater improvements for rTMS alone compared to CIMT alone (p=0.06). rTMS and CIMT effects did not appear synergistic. rTMS did not decrease function in children with ipsilateral CST projections (MA increased 5.1(4.6)%, p=0.04). Unaffected hand function did not decrease with rTMS. Procedures were well tolerated with no serious adverse events.

Conclusions: Non-invasive brain stimulation trials are feasible in children with perinatal stroke. Inhibitory, contralesional rTMS may enhance motor learning therapy in stroke-induced cerebral palsy. Pending long-term outcomes (2, 6 months) will help determine clinical relevance.

H7

Appropriate intervention and adequate dose: an implementation study for upper limb therapy for children with unilateral cerebral palsy

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Background/Objectives: Moderate to strong evidence exists for what constitutes adequate doses of goal-directed upper limb (UL) therapy using motor learning based approaches to improve UL skills for children with unilateral cerebral palsy (UCP). Despite this evidence, occupational therapists (OTs) generally fail to use a goal directed approach, persist in using therapies with limited evidence and ineffective models of therapy provision. This study aimed to develop and pilot the effectiveness of a multimodal research translation intervention to increase the appropriateness and dose adequacy of therapy for children with UCP.

Design: Before and after study.

Participants and Setting: Three teams of OTs participated. Team A (n=4) provided tertiary statewide services for children with CP; Team B (n=3) statewide pediatric rehabilitation services and Team C (n=3) regional hospital and community paediatric OT.

Methods: The translational intervention targeted five evidence criteria and comprised audit and feedback, barrier identification, and tailored interactive training. Medical chart audits were conducted prior to (n=43) and 12 months after (n=53) the intervention. Primary process outcomes related to evidence criteria included the proportion of children who had (1) goals set prior to therapy; (2) goals measured pre/post therapy; (3) received contemporary motor learning based approach (e.g. constraint therapy CIMT, bimanual therapy); (4) received an adequate dose (30h); and (5) measured UL outcomes pre/post therapy. Secondary patient outcomes were occupational performance (Canadian Occupational Performance Measure

COPM); and bimanual performance (Assisting Hand Assessment AHA).

Results: Teams engaged families in collaborative goal setting on 81% of occasions, increasing to 89% post intervention, with the greatest change for Team C (29%). Post intervention, there was increased measurement of client goals before (16%) and after (24%) therapy episodes. Prior to the intervention, CIMT was rarely used (7%) and this increased to 45% post intervention. Children receiving the target dose of therapy increased from 0 to 9.6%. Additional therapy dose was achieved through greater use of home programs and group delivered therapies. Measurement of UL outcomes pre/post therapy episodes increased by 19% and 23% respectively. Post intervention, fourteen children with completed pre/post assessments had a statistically, clinically meaningful change of 8.4, 95% CI 3.9, 13; p=0.002 AHA units. Twenty-one children (40%) with complete pre/post assessments achieved significant, clinically meaningful gains in COPM performance (MD 2.3, 95% CI 1.4, 3.1; p<0.001).

Conclusions: Modest changes in clinical practice behaviour, consistent with published research were achieved with an active multimodal research translation intervention. This intervention will now be more rigorously investigated in a cluster randomized trial.

H8

The effects of intense combined constraint and bimanual training on upper extremity functioning among children with hemiplegic cerebral palsy: does severity matter?

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Background/Objectives: Children with hemiplegic cerebral palsy (HCP) are limited in unimanual as well as bimanual function. The literature reflects less attention to the child's subjective experience during bimanual functions. The aim of the current study was to assess the influence of intensive combined, constraint and bimanual training program on upper extremity (UE) functions and to assess the influence of hand impairment severity on intervention outcome. In addition the child's experience on his abilities pre and post intervention were assessed. Design: Self control Study.

Participants and Setting: Twenty three children aged 6 to 11 years, 7 with Manual Ability Classification System (MACS) 1, 12 with MACS 2 and 6 with MACS 3, participated in the study.

Methods: A child-friendly, functional oriented program that daily combined an hour of constraint with 5 hours bimanual practice, for 2 weeks, 5 days per week. The data of the Assisting Hand Assessment (AHA) for bilateral UE activity, the Jebsen-Taylor Test of Hand Function (JTTHF) assessments for unimanual UE speed and function and the Children Hand-use Experience Questionnaire (CHEQ) were collected three times: prior to, immediately post and 3-months post- intervention.

Results: Significant differences in AHA and JTTHF scores were noted between groups based on MACS score. Children with MACS level 1 had significant better performance; AHA logits scores were 2.55 (1.2) among children with MACS 1, as opposed to 0.07 (1.2) and -1.92 (1.1) among children with MACS level 2 and 3 respectively (p<0.01). Significant improvements were noted in both the AHA and the JTTHF, immediately and 3-months post -intervention as compared to baseline in all outcome variables with no significant differences in improvement trend between MACS levels; AHA logits changed by 1.6 (0.4), 2.3 (1.1) and 1.4 (0.5), in MACS level 1,2 and 3 respectively. Although objective improvement was noted, no subjective change was noted in child's perspective on his ability in using the affected hand, with no influence according to severity levels.

Conclusions: The results of this study suggest that a combined constraint and bimanual intensive training program improves bimanual as well as unimanual hand function in different levels of upper extremity severity among children with hemiparetic cerebral palsy, without an effect on the child's perspective of improvement. Future studies should address the influence of the child's awareness following participation in such an intensive functional program, when using his hemiplegic upper extremity during bimanual function.

H9

Efficacy of a web-based multimodal therapy program on occupational performance, upper limb function, and visual perception for children with unilateral cerebral palsy

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Background/Objectives: To investigate the effectiveness of a web based multimodal program, 'Move it to improve it' (MitiiTM) delivered in the home environment compared to standard care for children with unilateral cerebral palsy (CP) on measures of activities of daily living (ADL), goal attainment, upper limb function and visual perception.

Design: Matched pairs randomized controlled trial.

Participants and Setting: Eighty-two children with spastic hemiplegia matched for age (mean age=11y 6mo [2y 5mo]; range 8–17y), sex (42 males) and MACS level (MACS I=16, II=66) were recruited at a tertiary referral centre.

Methods: Participants were randomly allocated to either the intervention group (Mitii™ program for 20 wks, 20–30 min daily) or the control group (standard care for 20 wks). The Mitii™ program consists of approximately 60% upper limb (UL) and visual perception/cognitive activities and 40% grossmotor activities. Outcomes measures were the: (1) Assessment of Motor and Process Skills (AMPS); (2) Assisting Hand Assessment (AHA); (3) Jebsen-Taylor Test of Hand Function (JTTHF); (4) Canadian Occupational Performance Measure (COPM); and (5) Test of Visual Perceptual Skills (non-motor)

3rd edition (TVPS). Data were compared between groups at baseline and 20 weeks using independent sample *t*-tests (SPSS v22)

Results: There were no differences at baseline between groups on any measure. Retention rates were 38/41 children (92.7%) in the intervention group and 35/41 children (85.4%) in the control group. Participants in the intervention group completed on average 60% of maximum dose (range 3.6-54.5h). The intervention group demonstrated significantly greater improvements on the AMPS Motor Scale (MD=0.34; 95% CI=0.14-0.53; p<0.001), AMPS Process Scale (MD=0.41, 95% CI=0.20–0.63; *p*<0.001), COPM Performance Scale (MD=1.32; 95% CI=0.58-2.05; p<0.001), COPM Satisfaction Scale (MD=1.55; 95% CI=0.75-2.34; p<0.001) and JTTHF for the dominant UL (MD=-6.69, 95%CI=1.29-12.09; p=0.02) compared to the control group. Secondary analysis of the JTTHF at 20 weeks by MACS level, showed that children in the intervention group classified as MACS I trended towards an improvement (MD=-44.6; 95%CI=-90.86-1.66; p=0.06). Analysis by domain of the TVPS showed a significant improvement in the intervention group in the Visual Spatial Relationships domain (MD=1.8; 95% CI=0.004-3.6; p=0.05) compared to the control group. There was no significant difference between groups at 20 weeks on the AHA (p=0.56).

Conclusions: The MittiTM program is an effective intervention for children with unilateral CP to improve ADL motor and processing skills, individual goal attainment and visual-spatial abilities. Children classified as MACS level II may require additional specific UL training to improve their manual abilities. The MittiTM program offers a home-based alternative to standard face-to-face therapy that can provide significant benefits for children with unilateral CP.

H10

Knowledge translation in constraint-induced movement therapy and hand-arm bimanual intensive therapy: clinicians' perspectives on barriers and facilitators for the uptake of intensive upper extremity training in children with hemiplegia in Canada

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Background/Objectives: Intensive upper extremity training programs (CIMT/HABIT) have high-level evidence supporting improved function in children with hemiplegia. However, implementation of these programs in the clinical context requires novel service delivery models, and may not be accessible across Canada. The objective of this study was to address this gap in knowledge uptake by identifying facilitators, barri-

ers and strategies to promote implementation of this treatment

Design: Qualitative design, content analysis.

Participants and Setting: Five focus groups were conducted across Canada (4/5) and 1 in the Netherlands, with a total of 43 participants from diverse professional backgrounds. Telephone surveys were carried out with managers of 27 pediatric rehabilitation centers in Canada.

Methods: Focus groups: Purposeful sample to include centers with and without established CIMT/HABIT programs, different language milieus, and participants representing community and rural settings, professions and administrative/clinical roles. Audio recordings were transcribed verbatim. Survey: Openended questions through telephone probed further views of uptake and implementation of evidence. Two coders iteratively and independently explored data from both methods through content analysis, using NVivo10. Coding identified surface meaning of participants accounts clustered initially into barriers and facilitators for evidence uptake, and further conceptualized into the aspects related to CIMT/HABIT programs that could be acted upon.

Results: Content explaining the phenomena of implementation related to: beliefs and values about evidence-based practice, opportunities for continuing education, proximity between researchers and clinicians, access to the literature, and the presence of a champion. Specific to CIMT were factors related to the pressure from parents and media questioning this intervention, and adaptation of evidence to clinical reality. Organizational aspects included the support and flexibility of families, having a critical mass of clients, being connected to a specialized school and managerial openness to new ideas and to restructure service provision (e.g. funds and personnel). Subjective stakeholders-related aspects related to maintaining or continuing a program included: parents enthusiasm and belief in the value of the program, staff motivation generated through initial success, and perceived benefits to the population being served.

Conclusions: Uptake and implementation of the evidence in intensive upper extremity programs require many steps described in the knowledge translation cycle, including identification of barriers and facilitators, which should be adapted to the local context. Factors identified in this study should be carefully considered in clinical settings, so as to facilitate the uptake of this effective treatment program. Consideration of families needs, identification of knowledge champions, facilitation of access to research evidence and creative program management are key elements to enable program development.

Free Papers I

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The effect of varying stiffness of an ankle-foot orthosis on walking ability in children with cerebral palsy

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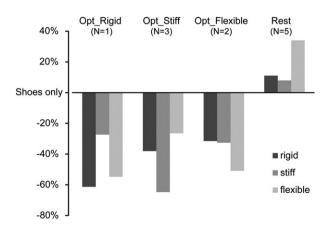
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Background/Objectives: Mobility limitations in children with spastic Cerebral Palsy (SCP), such as an increased walking energy cost (EC), may be associated with gait abnormalities. Gait characterized by excessive knee flexion in stance can be corrected with a rigid ventral shelf Ankle Foot Orthosis (AFO). However, this type of AFO inhibits push-off power. A more spring-like AFO may support push-off, therewith reducing walking EC. Studies in adults with neurological impairments showed that the reduction in walking EC could be optimized by choosing the correct AFO stiffness. This might also apply to children with SCP. This study aims to evaluate the effect of varying AFO stiffness on reducing walking EC in children with SCP.

Design: Repeated measures observational study.

Participants and Setting: Eleven SCP children, walking with excessive knee flexion in stance, participated (mean age: 10 [2], Gross Motor Function Classification System levels: I–III). Methods: Participants were prescribed with a hinged ventral shelf AFO (Neuro Swing®, Fior & Gentz, Germany), of which stiffness towards dorsiflexion was varied into three configurations: rigid (8.1Nm/deg), stiff (1.2Nm/deg) and flexible (0.5Nm/deg). After a 4-week accommodation period to each configuration (randomly applied), a rest test and 6-minute walk test at comfortable speed were performed to determine the net nondimensional (NN) EC [J/kg/m]. The NN-EC relative to speed-matched control cost was determined, referred to as SMC-EC [%]. For comparison, SMC-EC while walking with shoes only was also determined.

Results: The mean SMC-EC (SD) in the shoe-only condition was 258% (60%). Compared to shoe-only, six participants showed a reduction in their SMC-EC with all AFO configura-



tions (see Figure 1). In one participant, the rigid configuration accounted for the greatest reduction in SMC-EC (-61%), though at the expense of an 11% decrease in speed. In three other participants, the stiff configuration was most beneficial (SMC-EC: -65%, -64% and -65% and speed: -6%, -2% and +6%, resp.). In another two participants, the flexible configuration resulted in the greatest decrease in SMC-EC (-57% and -45%), at a concomitant increase in speed (+4 and +35%). In the remaining five participants, the AFO had (almost) no effect or a detrimental effect on SMC-EC (-6% to +87%). In two of them, gait speed concomitantly deteriorated (-6% to -27%), while in the other three, gait speed either remained unchanged or improved (+2% to +38%).

Conclusions: In children with SCP walking with excessive knee flexion, applying different degrees of AFO stiffness altered walking ability in most participants. Yet, the optimal AFO stiffness varied between participants. This emphasizes the importance of an individual approach to AFO prescription in these children. Further study will yield insight on how different AFO mechanics and patient characteristics contribute to reducing the EC while walking with AFOs in children with SCP.

The effect of different AFO stiffness configurations on the net non-dimensional energy cost (NN-EC) relative to speed matched control NN-EC (SMC-EC [%]), compared to walking with shoes only. Results are averaged for different subgroups, based on greatest reduction on SMC-EC. Opt_rigid=rigid configuration resulted in greatest reduction; Opt_Stiff=stiff configuration resulted in greatest reduction; Opt_Flexible=flexible configuration resulted in greatest reduction; Rest=all configurations had no effect or a detrimental effect.

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Influence of different ankle-foot orthosis types on crouch gait: a retrospective review using computerized gait analysis

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Background/Objectives: Crouch is a common gait pathology seen in gait analysis laboratories. Persistent crouch has been shown to contribute to reduced walking speed, knee pain, increased energy expenditure and reduced walking capacity. One common intervention utilized to treat crouch gait is an ankle-foot orthosis (AFO). However, little is known about the relative effectiveness of different AFO types on crouch gait. Design: Retrospective cohort study.

Participants and Setting: Data was collected in a computerized gait analysis laboratory of a regional rehabilitation hospital. One hundred fifty-two (152) limbs were identified as demonstrating crouch gait during barefoot walking for whom AFO data was also collected. Mean age of the patients was 11.3 years (Range 3.8–23.6y). Diagnoses included cerebral palsy (72%), myelomeningocele (20%), traumatic brain injury (5%) and other (3%). Methods: A search of our database of patients evaluated between January 2010 and November 2013 was conducted. Three

hundred three (303) AFO-wearing limbs were identified, of which 152 were deemed to demonstrate crouch gait. Crouch was operationally defined as minimum knee flexion during the stance phase (MKFS) above 14.5° (>2 standard deviations above normal). The various AFOs worn were classified as either solid (SAFO/53.3%), hinged (HAFO/20.4%), ground reaction (GRAFO/19.7%), dynamic (DAFO/3.9%) and posterior leaf spring (PLS/2.6%). PLS and DAFO data were not analyzed due to insufficient power. The difference in MKFS for each patient when barefoot versus braced was averaged for the 3 most common brace types, SAFOs, HAFOs and GRAFOs. A Pearson Chi-Square analysis was performed to compare the likelihood of improvement of crouch by the 3 major brace types. Those limbs in which MKFS decreased by ≥2.5° in the brace were defined as being 'improved,' while those limbs in which MKFS increased by ≥2.5° were defined as having 'deteriorated' (Those ±2.5° were regarded as unchanged.). A Chi square analysis was performed to determine significant differences between the 3 primary brace types (p<0.05).

Results: MKFS was decreased (crouch reduced) by an average of 8.90°in GRAFOs, by 4.19° in SAFOs and by only 0.40° in HAFOs. Chi-Square analysis revealed GRAFOs to be significantly more likely to improve crouch (86.7%) than were SAFOs (50.6%), and SAFOs significantly more likely to improve crouch than were HAFOs (32.3%), p<.001. Crouch gait was unchanged or worsened in 67.7% and 49.4% of limbs wearing a HAFOs or SAFOs respectively. Only 13.3% of GRAFO limbs showed no improvement in crouch.

Conclusions: GRAFOs appear to provide the best biomechanical constraint to excessive stance phase knee flexion (crouch), as these were easily the most reliable design assessed. SAFOs, while the most commonly prescribed, stand only a 50% chance of reducing crouch gait. Computerized gait analysis is an effective tool for the objective assessment of AFO efficacy.

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The effect of foot type on ankle power in children with cerebral palsy

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Background/Objectives: The relationship between foot type and foot function is not well established in pediatric or adult population. However, in individuals with Cerebral Palsy (CP), foot type may play a significant role in function. Deficient selective motor control (SMC) and altered muscle balance can prevent adequate compensation for foot deformity. The purpose of this study was to examine the relationship between foot type and ankle power in ambulatory children with CP. Design: Retrospective.

Participants and Setting: Patients with CP (5-18y) who completed a 3-D gait analysis at our institution.

Methods: Charts were reviewed to identify children meeting the inclusion criteria: diagnosis of CP, GMFCS I/II, and no prior surgery. Exclusion criteria: >5° ankle equinus and/or

>10° knee or hip flexion contracture in any limb, prior rhizotomy or limb surgery (bony or soft tissue), and Botox injections in the 6 months prior to testing. The uninvolved limb of hemiplegic individuals was excluded. Review included demographics, physical exam, and plantar pressures. Foot type (planovalgus, cavovarus, rectus) was defined by ankle exam, observed static stance posterior video view, and coronal index. Coronal index (Fig 1) was recorded from dynamic plantar pressures according to Miller et al. JPO, 2002. Once foot type was determined, it was correlated to kinetics. An average of at least 3 representative gait cycles was calculated to determine the sagittal ankle power (3rd rocker) and maximum stance ankle moments calculated in OrthoTrak. The inter-rater reliability of two raters defining foot type was assessed by calculating weighted Kappa statistic and 95% CI. Relationships between continuous outcomes and categorical predictors were assessed with single-factor ANOVAs, while associations between categorical variables were tested with Chi Squared or Fisher's exact tests.

Results: Of 45 participants, (N=55 feet) met the inclusion criteria: planovalgus (N=30), cavovarus (N=15), rectus (N=10). Inter-rater reliability of defining foot type was good (weighted Kappa 0.71, 95% CI 0.43–1.0). Planovalgus feet had a trend towards greater power generation during stance than cavovarus feet (p=0.057, Table 1). Increased selective motor control (SMC) (p=0.010) and GMFCS I(p=0.012, Table 2) were associated with increased ankle power. Ankle moments were not different between foot types.

Conclusions: Among children with CP, those with planovalgus feet tend to produce more sagittal ankle power than those with cavovarus feet. We postulate that children with cavovarus feet may have greater muscle imbalance and neurological impairment affecting the foot; this is consistent with the association of increased power in participants with better SMC. Ours is the first study to demonstrate that SMC and GMFCS level correlate with ankle power generation. This is the largest (N=55) study on the effect of foot type on function in children with CP prior to surgical intervention. Future studies would be useful to assess changes in powers after surgical foot realignment in individuals with CP.

Table 1 Foot type and power

Foot Type	Number of limbs (N)	Ankle Power (mean <u>+</u> sd) Watts/kg	Between groups comparison (p-value)		
Planovalgus	30	1.21 +0.47	0.057		
Cavovarus	15	0.87 ±0.32	g aller on		
Rectus	10	1.21+0.50			

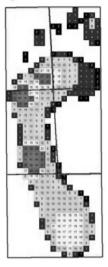
Table 2 Ankle power and GMFCS

GMFCS level	Number of limbs (N)	Ankle Power (mean <u>+</u> sd) Watts/kg	Between groups comparison (p-value)
GMFCS level I	27	1.27 + 0.42	0.012
GMFCS level II	26	0.96+0.46	1.7.1807.

Table 3 Powers for individuals with Diplegic vs Hemiplegic CP

Type of CP	Number of limbs (N)	Ankle Power (mean <u>+</u> sd) Watts/kg	Between groups comparison (p-value)
Diplegic CP	23	1.22 + 0.56	0.128
Hemiplegic CP	32	1.03 ± 0.36	, 1 Still Held .

Figure 1



Foot is bisected by a line crossing from heel center to space between 2nd and 3rd metatarsal heads. The foot is then divided to 3 equal thirds along its length, with heel treated as one segment.

Coronal Index shows the impulse distribution in the foot:

(medial midfoot + medial forefoot) – (lateral midfoot + lateral forefoot)

Sum of lateral and medial foot pressure

Rectus (straight) feet tend to have values between -30 and 12, Planovalgus feet (high medial impulse) have values >12 and Cavovarus feet (high lateral impulse) have values < -30 ^{1-3.}

Figure 1: Coronal Index

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The influence of the unaffected hip on gait kinematics in patients with hemiplegic cerebral palsy

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Background/Objectives: Patients with hemiplegic cerebral palsy (HCP) have transverse plane gait deviations that may include the contralateral 'uninvolved' side. Are these compensations or primary abnormalities? The aim of this study is to quantify the static rotational profile, the dynamic position during gait and determine if any correlations between the involved and uninvolved side exist.

Design: Retrospective Analytic Observational Cohort study design of children with a diagnosis of HCP evaluated at the Shriners Hospital for Children, Portland. Clinical and gait measurements were analyzed and compared within the cohort and to a population of typically-developing (TD) children.

Participants and Setting: Inclusion Criteria: Diagnosis of HCP Patient age >3 years and <19 years Gross Motor Function Classification System level I or II Complete clinical and Motion Lab evaluation No prior history of bony surgery.

Methods: Static prone internal and external rotation of the hip, stance phase dynamic rotation of the hip and pelvis, knee progression angle* (mean hip rotation + mean pelvic rotation) were analyzed. Statistical analysis with unpaired t-test (significance level set at 0.05) * van der Linden et al, DMCN 2007.

Results: All 171 eligible patients participated in this study. See attached Tables 1 and 2 for significant results.

Conclusions: As expected, children with HCP displayed abnormalities of the affected limb and pelvis which positively correlated to the static internal rotation of the affected hip. Children with less than 66% internal rotation of the total arc of motion did not display any further significant differences on the affected side apart from mild pelvic retraction, and the affected knee progression angle was normal. However, children with greater than 66% internal rotation exhibited extensive abnormalities including dynamic pelvic retraction, dynamic internal

Table 1: Children with <66% vs >66% Internal Rotation of Total Arc of Motion in the Affected Hip

	TD (control)	H Affec	>66% VS <66% (p -value)	
		<66%	>66%	
Hip Internal Rotation (static)	Rotation (static) 52 ± 10		69 ± 9	.0001
Pelvic Rotation (dynamic)	2 ± 2	Retracted 5 ± 6	Retracted 8 ± 7	.0056
Hip Rotation (dynamic)	-3 ± 9	-2 ± 9	-13 ± 11	.0001
Knee Progression Angle	-1 ± 9	1 ± 10	- 8 ± 9	.0001

Table 2: Children with >66% Internal Rotation of Total Arc of Motion in the Affected Hip

	TD (control)	HCP - Affected limb	Contralate	>66% VS <66% (p-value)	
			>66% IR (n=23)	<66% (n=77)	
Hip Internal Rotation (static)	52 ± 10	69 ± 9	65 ± 10	50.3 ± 9	.0001
Pelvic Rotation (dynamic)	2 ± 2	Retracted 8 ± 7	Protracted 5.1 ± 6	Protracted 8.7 ± 7	.027
Hip Rotation (dynamic)	-3 ± 9	-13 ± 11	-3.5 ± 8	3 ± 7	.0004
Knee Progression Angle (dynamic)	-1 ± 9	-8±9	-8.6 ± 7	-5.7 ± 6	.048

hip rotation and dynamic internal knee progression. The extent of gait abnormalities of the contralateral limb was similarly related to the static internal rotation of the hip. Children with >66% internal rotation had greater dynamic internal hip rotation and internal knee progression angle. Children with <66% internal rotation of the unaffected hip had dynamic external rotation of the hip but still had a negative knee progression angle. In summary, the 'unaffected' side in patients with HCP influenced gait kinematics. If static internal hip rotation exceeds 66% of the total arc of motion, almost all studied static and gait parameters were abnormal in HCP children, regardless if it was the affected side. Compensations on the "unaffected" side appear to be somewhat limited if the anatomic alignment is significantly asymmetric. This may be one reason transverse plane pelvic changes after femoral rotation osteotomy are unpredictable. The influence of contralateral hip anatomic alignment on the outcome of femoral rotational osteotomy will be analyzed in the future.

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Kinematic profiles of Gross Motor Function Classification System level III functional subaroups

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Background/Objectives: Previous work suggests that only 1/3 of children with cerebral palsy (CP) at Gross Motor Function Classification System (GMFCS) III can voluntarily alter the velocity of their gait.1 Clinicians have used this ability to control gait velocity as an indicator of relatively less motor impairment within GMFCS III participants, which in turn has guided surgical management of gait deviations. We hypothesize that the kinematic profile, as reflected by the gait deviation index (GDI)² discriminates between participants at GMFCS III who can/cannot control their gait velocity.

Design: Retrospective convenience cohort.

Participants and Setting: Of 103 participants with diplegic CP, age range 5 to 20.8 years, referred for gait studies in a specialty pediatric hospital.

Methods: GMFCS levels and demographic data were recorded. Temporo-spatial and kinematic parameters were obtained at self-selected free and fast gait speeds (previously reported).^{1,3} Participants were categorized by GMFCS level and ability to

Table 1

Comparisons	Gait Deviation	Perry	
	Index	Segment	Locomotor
			Segment
I vs II	I: 80.3	n/a	n/a
	II: 74.2		
	p=0.214		
I vs III	I: 80.3	III: ↑Trunk Flex	III: ↑ Hip Flex St
	III: 64.4	(p<0.000)	(p<0.000)
	p=0.000		↑ Knee Flex St
	•		(p<0.000)
			↓ Knee Range
			Sw
			(P<0.000)
II vs III	II: 74.2	III: ↑Trunk Flex	III: ↑ Knee Flex
	III: 64.4	(p<0.000)	St
	p=0.010	(F)	(p=0.020)
	F		(4)
I vs IIIA	I: 80.3	n/a	n/a
	IIIA: 70.2		
	p=0.080		
I vs IIIB	I: 80.3	III: ↑Trunk Flex	III: ↑ Hip Flex St
	IIIB: 60.0	(p<0.000)	(p<0.000)
	p=0.000	(F)	↑ Knee Flex St
	F		(p<0.000)
			↑ Knee Flex Sw
			(p=0.016)
			↓ Knee Range
			Sw
			(p=0.002)
II vs IIIA	II: 74.2	n/a	n/a
	IIIA: 70.2		
	P=1.0		
II vs IIIB	II: 74.2	III: ↑Trunk Flex	III: ↑ Hip Flex St
	IIIB: 60.0	(p<0.000)	(p=0.020)
	p=0.001	, ,	↑ Knee Flex St
			(p=0.001)
			↑ Knee Flex Sw
			(p=0.006)
IIIA vs IIIB	IIIA:70.2	No	IIIB:
	IIIB: 60.0		↑ Knee Flex Sw
	p=0.014		(p=0.025)
	F		<u> </u>

St = stance phase; Sw = swing phase; n/a = not applicable

increase gait velocity (GMFCS I=33, II=35, III=35, IIIA [able to increase speed]=15, IIIB [unable to increase speed]=20 participants). Kinematic profiles for each group were calculated by the GDI. 23 additional kinematic parameters (6 passenger, 17 locomotor segments) were analyzed.⁴ Analysis of variance by GMFCS level was calculated for the GDI and the 23 kinematic parameters (p<0.05).

Results: GDI was significantly different between GMFCS I versus III, II versus III, I versus IIIB, II versus IIIB, and IIIA versus IIIB, with the lower functional GMFCS levels showing greater deviation in GDI for all comparisons.(Table 1) In those comparisons with differences in GDI, the significant passenger segment deviation was increased trunk flexion; the significant locomotor segment deviations were increased hip/ knee flexion in stance phase, and increased knee flexion/ diminished dynamic range in swing phase.

Conclusions: Clinicians recognize that impairment changes dramatically between GMFCS I/II versus III, and that the level of impairment within GMFCS III has a wide range. GDI by GMFCS supports these clinical observations. Subdivision of GMFCS III based upon ability to voluntarily control gait speed was also supported by the GDI, which revealed that the IIIA participants had kinematic indexes comparable to I/II groups, while IIIB group had indexes that deviated significantly from higher functioning groups. This supports the clinical practice of interpreting the ability to control gait velocity as an indicator of relatively less motor impairment within GMFCS III participants. Future research will focus on identifying additional discriminatory variables for GMFCS IIIA versus IIIB.

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Complexity of neuromuscular control is reduced during gait among individuals with cerebral palsy

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Background/Objectives: One theory for how humans control movement is that muscles are activated in weighted groups called 'synergies'. Synergies are theorized to provide a simplified neuromuscular control strategy compared to controlling each muscle individually. Recent studies have found that synergies are altered after stroke, and that the structure of synergies is related to severity and time after injury. The aim of this research was to determine if synergies are altered in children with CP, and to evaluate the relationship between synergy structure and clinical measures of functional ability, selective motor control, and energy costs of walking.

Design: Retrospective cross-sectional.

Participants and Setting: Individuals who had received motion analysis at a single clinical center prior to February 2013 were included in this study, resulting in 549 children with CP and 84 typically-developing children (Table 1).

Methods: Electromyography (EMG) was collected during gait from the rectus femoris, medial and lateral hamstrings, gas-

Table 1: Subject cl	haracteristics
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	Age	Height	Mass	Speed	GM	FCS	, % 0	fsut	jects	F	AQ,	% (of su	bjec	:ts
N	years	meters	kilograms	meters/sec	1	II	Ш	IV	NA	< 6	7	8	9	10	NA
84	10.4 ± 3.5	1.43 ± 0.21	40.0 ± 16.3	0.97 ± 0.25											-
122	12.9 ± 8.1	1.42 ± 0.19	40.3 ± 18.3	1.03 ± 0.23	52	13	1	0	34	3	3	14	41	35	3
266	10.9 ± 7.3	1.32 ± 0.20	33.6 ± 16.8	0.88 ± 0.25	29	27	11	0	33	11	10	24	39	14	1
73	10.8 ± 6.3	1.32 ± 0.20	32.5 ± 15.0	0.77 ± 0.30	12	30	19	5	33	14	19	22	34	8	3
88	11.5 ± 7.2	1.32 ± 0.19	32.1 ± 14.6	0.55 ± 0.28	0	12	38	10	40	55	11	18	11	2	2
	84 122 266 73	N years 84 10.4 ± 3.5 122 12.9 ± 8.1 266 10.9 ± 7.3 73 10.8 ± 6.3	N years meters 84 10.4 ± 3.5 1.43 ± 0.21 122 12.9 ± 8.1 1.42 ± 0.19 266 10.9 ± 7.3 1.32 ± 0.20 73 10.8 ± 6.3 1.32 ± 0.20	N years meters kilograms 84 10.4 ± 3.5 1.43 ± 0.21 40.0 ± 16.3 122 12.9 ± 8.1 1.42 ± 0.19 40.3 ± 18.3 266 10.9 ± 7.3 1.32 ± 0.20 33.6 ± 16.8 73 10.8 ± 6.3 1.32 ± 0.20 32.5 ± 15.0	N years meters kilograms meters/sec 84 1.04 ± 3.5 1.43 ± 0.21 40.0 ± 16.3 0.97 ± 0.25 122 12.9 ± 8.1 1.42 ± 0.19 40.3 ± 16.3 1.03 ± 0.23 266 10.9 ± 7.3 1.32 ± 0.20 33.6 ± 16.8 0.88 ± 0.25 73 1.8 ± 6.3 1.32 ± 0.20 3.5 ± 15.0 0.77 ± 0.30	N years meters kilograms meters/sec 1 84 1.04 ± 3.5 1.43 ± 0.21 40.0 ± 16.3 0.97 ± 0.25 122 12.9 ± 8.1 1.42 ± 0.19 40.3 ± 18.3 1.03 ± 0.23 52 266 10.9 ± 7.3 1.32 ± 0.20 33.6 ± 16.8 0.88 ± 0.25 92 73 10.8 ± 6.3 1.32 ± 0.20 3.5 ± 15.0 0.77 ± 0.30 12	N years meters kilograms deters/se 1 I I 84 10.4 ± 3.5 1.43 ± 0.21 40.0 ± 1.63 0.97 ± 0.25 122 12.9 ± 8.1 1.42 ± 0.19 40.3 ± 1.83 1.03 ± 0.23 2.2 1.2 2.0 2.5 2.5 2.7 2.7 3.3 ± 6.16.8 0.88 ± 0.25 2.9 2.7 2.7 3.0 ± 6.3 3.2 ± 0.20 3.2.5 ± 15.0 0.77 ± 0.30 1.2 3.0	N years meters kilograms meters/se I II III 84 10.4 ± 3.5 1.43 ± 0.21 40.0 ± 16.3 0.97 ± 0.25	N years meters kilograms meters/ears 0.1 1 II IV 84 1.04 ± 3.5 1.43 ± 0.21 40.0 ± 1.63 0.97 ± 0.22	N years meters kilograms meters/ears 0.1 I II IV NA 84 1.04 ± 3.5 1.43 ± 0.21 40.0 ± 16.3 0.97 ± 0.22	N years meters kilograms meters/ears 0.1 II IV NA •	N years meters kilograms meters/sec 1 I II IV NA < 6 7 84 10.4 ± 3.5 1.43 ± 0.21 40.0 ± 1.63 0.97 ± 0.25 -	N N N N N N N N N N	N years meters kilograms meters/ears 1 I II IV NA < 6 7 8 9 84 1.04 ± 3.5 1.43 ± 0.21 40.0 ± 16.3 0.97 ± 0.25	N years meters kilograms meters/ears 1 II II IV NA < 6 7 8 9 1 84 1.04 ± 3.5 1.43 ± 0.21 40.0 ± 16.3 0.97 ± 0.25

GMFCS: Gross Motor Functional Classification FAQ: Functional Activity Questionnaire

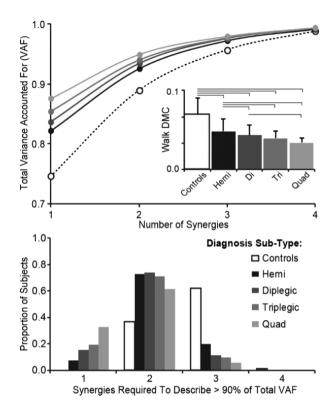


Figure 1: Control complexity during gait, as measured by synergies, was significantly reduced among children with CP compared to typically-developing children. (Top) Total variance accounted for (VAF) by one to four synergies was significantly greater for children with CP and increased with severity of diagnosis sub-type. (Top, inset) The Dynamic Motor Control index (Walk DMC), calculated as one minus the average VAF, decreased with severity of diagnosis sub-type. Brackets represent significant difference (p<0.05). (Bottom) Fewer synergies were required to describe greater than 90% of the total VAF among children with CP than typically-developing children.

trocnemius, and soleus while participants walked at their self-selected speeds. Synergies were calculated from EMG over one gait cycle for each participant using nonnegative matrix factorization. This algorithm calculates, for a given number of synergies, muscles that are commonly activated together and the activation of each synergy over the gait cycle. Complexity of control was evaluated by the variance in EMG accounted for (VAF) by groups of one to four synergies.

Results: Children with CP required fewer synergies to describe variance in EMG activity compared to typically-developing children (Figure 1). The synergies of the

typically-developing children were similar to previous studies and were consistent across walking speeds. For the typically-developing children, 3 or more synergies were required to describe greater than 90% of the variance in EMG for 63% of the participants. However, for the children with CP, only 1 or 2 synergies were required to describe greater than 90% of the variance in EMG for 88% of the participants. VAF increased with severity of cerebral palsy as measured by both GMFCS and FAQ levels. Complexity of synergies was also related to selective motor control (r=0.44), strength (r=0.48), spasticity (r=-0.33), and energy costs of walking (r=-0.44) such that individuals with simpler control had worse selective motor control, less strength, more spasticity, and higher energy costs.

Conclusions: Children with CP utilized fewer synergies during gait than typically-developing children and the structure of synergies were similar to adult stroke survivors. These results suggest that similar changes in neuromuscular control are adopted after brain injury. Measures of synergies during gait and other activities may provide a new metric that can be used to develop rehabilitation strategies and evaluate the impact of treatment for individuals with CP.

I7 Postural control exercise interventions for children with cerebral palsy: a systematic review

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Background/Objectives: Despite cerebral palsy (CP) being defined as a 'disorder of posture and movement', the development of successful interventions to treat postural dysfunction is in its infancy. This systematic review aimed to identify postural control interventions for children with CP, to critique their efficacy, and to make recommendations for improved therapeutic management of this fundamental attribute of CP. Design: Systematic review.

Participants and Setting:

Methods: Six electronic databases were searched for population ('cerebral palsy' OR 'brain injury') and intervention type ((postur* OR balance OR 'Postural Balance' (MeSH)) AND ('intervention' OR 'therapy' OR 'exercise' OR 'treatment')). Secondary searches included reference list checking, searches of intervention names and authors, and citation tracking. Included papers (1) were full articles published after 1980 in English, and (2) included 0 to 18 year old children with CP, (3) reported land based postural control exercise intervention/s, and (4) included outcome measures for postural control (stability or orientation). Papers were excluded if they reported (1) passive equipment interventions (e.g. seating or support garments), (2) water-based interventions, (3) medical or surgical interventions, (4) general gross motor measures that did not specifically evaluate postural control, or (5) non-systematic reviews. Included articles were evaluated according to level of evidence and conduct as recommended by the AACPDM guidelines (2008).

Results: Of 45 articles met criteria for evaluation, including 12 intervention types purported to impact postural control or balance for children with CP. Ten intervention types appear to improve postural control and 2 do not. Moderate evidence was presented for five approaches: hippotherapy, treadmill training without body weight support, gross motor task training, reactive balance training and trunk strengthening on a vibration platform. Weak or conflicting evidence was presented for five interventions: hippotherapy simulators, virtual reality, visual biofeedback, treadmill training with body weight support and robotic assist (Lokomat) and FES. Interventions with no positive impact were: upper limb interventions and strength training. Interventions showing improvements were all reported in studies involving ambulant children with CP (GMFCS I-III).

Conclusions: Need for better postural control interventions for children with CP is gaining attention. Several potentially effective interventions have been identified; others that are not and some that require further investigation. Research is required to (1) establish responsive and reliable outcome measures (2) establish effective dose and assessment guidelines for existing interventions, (3) further evaluate newer interventions such as virtual reality and (4) to explore mainstream interventions that have demonstrated effectiveness for adults with brain injury, such as Pilates, yoga and Tai Chi.

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Lower limb muscle fascicle function during gait in young adults with cerebral palsy

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Background/Objectives: During gait in typically developed (TD) individuals, the muscles of the calf contract at levels that maintain a relatively constant muscle fibre length while the more compliant Achilles tendon acts like a spring to absorb and return energy during each step. However, in response to spasticity, the calf muscles of individuals with Cerebral Palsy (CP) become smaller, weaker, more resistant to stretch, have increased antagonist co-contraction and the Achilles tendon becomes longer. These neuromuscular adaptations of the calf in CP could potentially reduce muscular forces and contribute to altered gait patterns, however it is difficult to know what impact these adaptations will have on fibre or tendon behaviour. The primary aim of this study was to investigate the function of lateral gastrocnemius muscle-tendon unit (LG_{mtu}) and fascicles (LG_{fas}) in young adults with CP compared to TD individuals. Design: Prospective case/control comparison.

Participants and Setting: Seven young adults with CP, age 15 to 21 years, (5 males, 2 female, 5 hemiplegia, 2 diplegia, GMFCS I=7) and 7 TD, age 17 to 22 years, participated in the study.

Methods: Participants walked at a self-selected speed on an instrumented treadmill while 3D kinematics and kinetics and ultrasound images of LG_{fas} were acquired. LG_{mtu} was determined based on a kinematic model. Primary outcome measures were changes in LG_{mtu} and LG_{fas} length and velocity during mid- and terminal-phase of stance. A two-tailed

Mann–Whitney U-test (p<0.05) was used to compare differences between groups.

Results: In mid-stance the LG_{mtu} increased length similarly in both groups (CP=10.2(3.8)mm; TD = 12.5(3.8)mm, p=0.41), however fascicles underwent significantly different length changes between groups (p<0.01) with CP LG_{fas} lengthening by 1.2(0.8)mm and TD LG_{fas} shortening by 1.2(1.2)mm. During terminal-stance the LG_{mtu} shortened less in CP group (CP=26.2(8.2)mm; TD=44.5(6.7)mm, p=0.01) and the LG_{fas} also shortened less (p=0.01) and at slower speeds (p=0.01) in the CP group (CP=1.5(1.3)mm and 6.2(5.2)mm/s; TD=5.0 (2.2)mm and 19.9(11.1)mm/s).

Conclusions: During walking, while the LG_{mtu} lengthens comparably between groups, the LG_{fas} in the CP group actually lengthens through mid-stance while they shorten in TD individuals. The eccentric action of the CP muscle fascicles during walking is consistent with the greater passive stiffness and perhaps greater reliance on passive rather than active muscle force. During the "push-off" phase the CP group had less LG_{mtu} and LG_{fas} shortening; thereby the muscle contraction contributes less to forward propulsion power during walking.

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Postural responses to visual and somatosensory perturbations in adults with cerebral palsy

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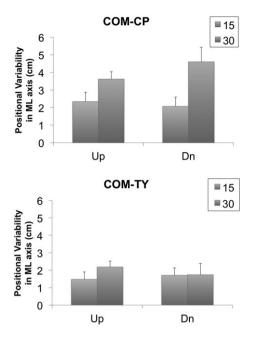
Background/Objectives: Deterioration of balance control has been found in adolescents and adults with Cerebral Palsy (CP) with aging. While musculoskeletal and sensory deficits may both affect balance, very little is known about how the sensory components play a role in balance control among individuals with CP. The current study is aimed at identifying postural responses to changes of the visual environment and the supporting surface in adults with CP in comparison to adults with typical development (TY).

Design: This is a cross-sectional study.

Participants and Setting: Five adults with spastic CP and seven adults with TY from the greater Philadelphia area volunteered in this study at Temple University.

Methods: Participants were instructed to stand on a platform in an immersive virtual environment consisted with three walls. The platform was either tilted up into dorsiflexion by 3°, or kept stationary. During the tilting condition, the platform was held at tilted position for the first 30 s, and then gradually returned to a neutral position over 30 seconds. The visual scene was set to either pitch up or pitch down, at either 15°/seconds or 30°/seconds. Center of mass (COM) throughout the trials was measured for analysis. Repeated measure ANOVAs were used to test the postural response dipected by COM as a function of Direction (Pitch Up or Pitch Down), Speed (15 or 30°/seconds), Floor (Tilted or Fixed), and Group (CP or TY).

Results: Greater speed of the visual scene movement induced greater positional variability of COM in both the anterior-posterior (AP; *p*<0.001) and the medial-lateral (ML; *p*=0.002) directions for both groups. The speed induced an even greater response in the ML axis in adults with CP in comparison to



adults with TY, as depicted by an interaction effect between group and speed (p=0.018). Furthermore, an exaggerated visual illusion that emerged from opposite movements of the visual scene and the platform turned out to impose greater postural response of adults with CP in the ML direction (p=0.023). There was no effect of platform movement in either group.

Conclusions: Overall, the results from the current study indicated that balance of individuals with CP was greatly affected by the visual scene movement. The reliance on visual information may indicate the potential deficits in the vestibular and/or somatosensory systems. The inability to compensate for changes in visual environment may hamper the maintenance of functional activity such as locomotion. Future studies are needed to investigate the engagement of vestibular and/or somatosensory systems in balance control in this population to further develop effective interventions.

Figure 1: Positional variability of COM in CP and TY during Pitch Up and Pitch Down conditions.

I10

What evidence exists on the effectiveness of the use of Robotic-Assisted Gait Training in children with neurological gait disorders?

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Background/Objectives: Refinements in robotic technology in recent years has seen several models of robotic-assisted gait trainer used in paediatric rehabilitation settings. While a strong body of evidence exists for adults with neurological impairments, less is documented about its potential in paediatric rehabilitation. We examine the efficacy of Robotic-Assisted Gait Training (RAGT) in improving the gait of children with

neurological gait disorders, from existing experimental studies, and in doing so explore other factors (e.g. degree of active participation and outcomes in relation to ICF domains) of the clinical application of RAGT in children.

Design: Systematic review.

Participants and Setting: Of 305 participants (45 control participants) were recruited by study authors. Neurological gait disorders included (but were not limited to) cerebral palsy (GMFCS I-V), myelomeningocoele, Guillian-Barré Syndrome and traumatic brain injury.

Methods: Data was extracted from experimental studies which met inclusion criteria (*n*=14), influenced by the AACPDM and PRISMA methods for developing systematic reviews. All studies underwent critical appraisal. Four RCTs were identified, with the remainder of studies before-after, cohort, case series or case study methodologies. Methodological heterogeneity prevented meta-analytic synthesis of data. This review aligned research outcomes with the International Classification of Functioning, Disability and Health (ICF) to inform directions for future research and enhance applicability of findings.

Results: Data included walking endurance, speed, functional gross motor performance, clinical gait analysis and some specific physiological outcomes as ways of determining RAGT clinical effectiveness. Most studies (n=9) cited between 2 and 5 RAGT sessions per week, over a 2 to 6 week testing period, and 5 studies reported outcomes on a single intervention session. RAGT appeared most effective when children were provided with additional extrinsic feedback during intervention (e.g. virtual reality games) and their functional gross motor performance shows moderate-level limitations in standing and walking (e.g. Gross Motor Function Measure-66: items D and E). There is some evidence that RAGT improves temporospatial and kinematic parameters of gait in children with CP following clinical gait analysis, however available data is limited. Researchers most frequently investigated RAGT outcomes in relation to activity limitations.

Conclusions: Results infer RAGT has functional, temporospatial and kinematic gait performance benefits for children with neurological gait disorders, although the evidence remains limited. This systematic review also investigates documented training parameters and contextualises findings back to the ICF framework.

Free Papers J

J1

Assessment of general movements by Prechtl Analysis and follow-up results in high-risk infants during the early intervention period

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Background/Objectives: This study was planned to assess general movements (GMs) by Prechtl Analysis and represent follow up results in high risk infants during the early intervention period of the first year of life.

Design: Double blind crossover study design.

Participants and Setting: The study included 28 preterm and low birth weight, high risk infants and were assessed by neonatologist and physiotherapist.

Methods: Assessment of neonatologist included demographic characteristics, neurological examination at 40 weeks, 3, 6, 12 months. Assessment of physiotherapist included 2 to 5 times of video recordings of GMs by Prechtl Analysis in between preterm to postterm 20th week of corrected age. In addition, physiotherapist assessed motor development in 3, 6, 12 months with Harris Neuromotor Test as well as with Gross Motor Function Measurement (GMFM) in 12 months. All infants underwent early physiotherapy programme. Agreement in assessments of neonatologist and physiotherapist was analysed by Cohen Adjusted Kappa (PABAK). To calculate the correlation, Spearman correlation rank was used.

Results: The retrospective analysis indicated that only 7.2% of infants had pathological ultrasound results. Only one infant had suspected of CP at the 40th week assessment while no infants had the diagnosis in the other follow ups. In GMs results, 17 infant showed abnormal movement in preterm and writhing movement and 15 of these infants had typical fidgety movements and considered as having typical neurological outcome. Only two infants had abnormal results from GMs. Cranial ultrasound, neurological assessment and GMs results were compared for the agreement. Perfect agreement was found between cranial ultrasound and GMs (adjusted κ (PA-BAK)=0.86). Neurological assessment and GMs had important agreement(adjusted κ (PABAK)=0.78). Strong correlation was found between: Apgar 5th minute score-respiratory distress syndrome, gestational age-birth weight, Apgar 5th minute score-gestational age, Gross Motor Function Measurement (GMFM) total score-Harris Neuromotor Test in 12th months, GMFM total score-walking age, walking age-birth weight (p<0.01). All the infants gained independent walking and mean age for the independent walking was 12.22 (2.08) months of corrected age. One of the two infant who had abnormal GMs died and the other had independent walking at the corrected age of 18 months and had minor neurological dysfunction.

Conclusions: Prechtl Analysis is important to detect not only neurological dysfunction but also typical neurological outcome in high risk infants during early intervention period, can be used complementary to other diagnostic and imaging techniques as well as one of the necessary assessment tools of early intervention.

J2 Supporting Play, Exploration, & Early Development of Infants born preterm (SPEEDI): a pilot randomized clinical trial

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Background/Objectives: Infants born prematurely are at increased risk for developmental disabilities. While federally mandated

Early Intervention (EI) is designed to help parents of infants with or at risk of developmental delays, services typically do not begin for months after hospital discharge, if at all.

Design: The purpose of this pilot randomized clinical trial study was to determine the feasibility of a novel intervention and collect pilot efficacy data.

Participants and Setting: Ten infants born preterm with a mean gestational age of 28.9 weeks were enrolled.

Methods: Infants were randomly assigned to the usual care or intervention group at 35 weeks post menstrual age. The usual care group received services typical to this NICU including referral to EI if eligible. The intervention group received a minimum of 10 therapist provided physical therapy sessions and 2 parent education sessions before hospital discharge and daily home activities provided by the parent after discharge. The home activities were reviewed and updated every 2 weeks by the therapist through 3 months of adjusted age. Descriptive statistics were used to document the feasibility of using SPEEDI during the transition from the NICU to home. Motor assessments using the Test of Infant Motor Performance (TIMP) were completed at baseline, hospital discharge and the end of the intervention. The Early Problem Solving Indicator (EPSI) and duration of midline hand behaviors were assessed at the end of the intervention. A blinded assessor completed all study assessments and reliability was greater than 90% on all outcome measures. Cohen's d was calculated to estimate the effect sizes for each of the 3 variables.

Results: One infant was lost to follow-up in each group. Parents in the intervention group documented that intervention activities were completed 67 to 130% (average 100%) of the recommended frequency at home. The infant who only completed 67% of the required intervention sessions was excluded from analysis as she did not meet the minimum frequency set a priori. At the end of the intervention period, the intervention group has a mean TIMP z-score of 0.75 (SD=0.10, n=2) and the usual care group had a mean TIMP z-score of -0.08 (SD=0.74, n=4). A Cohen's d of 1.291 suggests a large effect size for TIMP z-scores. The intervention group exhibited problem solving behaviors during 182.43 second (SD=30.51, n=2) and the usual care group only displayed problem solving behaviors during 122.33 seconds (SD=19.07, n=4). A Cohen's d of 2.673 suggests a large effect size. The intervention group had a mean duration of hands in midline of 158.08 seconds (SD=111.98, n=2) and the usual care group had a mean duration of hands in midline of 69.55 seconds (SD=56.61, n=4). A Cohen's d of 1.19 suggests a large effect size for hands in midline.

Conclusions: This study demonstrates parent ability to participate in a hospital to home intervention and preliminary efficacy of SPEEDI to improve motor, problem solving, and midline hand behaviors.

J3

Identification of motor disabilities using the Bayley Scales of Infant-Toddler Development-3rd edition (BSITD-3) and the Neuro-Sensory Motor Developmental Assessment (NSMDA) in a high risk infant follow-up program

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Background/Objectives: The purpose of this study was to compare the use of the BSITD-3 and the NSMDA in identifying infants with motor delays eligible for enrollment in early intervention services. The gold standard developmental test in the United States is the Bayley Scales of Infant Toddler Development-3rd edition, a norm-referenced test that evaluates acquisition of gross and fine motor milestones. The BSITD-3 has been criticized for overestimating developmental milestones and decreasing the number of infants eligible for early intervention. The NSMDA is designed to qualitatively assess defined aspects of motor development and associated systems and has only been used more recently in the United States. It is administered by physical therapists and scored using age-normed criteria along with functional grading of performance that can be used to identify infants with functional deficits that would benefit from treatment.

Design: Prospective cohort study.

Participants and Setting: Twenty-nine high-risk children were evaluated in the follow up clinic. The mean birth weight was 2438 (1200) grams and mean gestational age was 34.1 (5.9) weeks. Neonatal risk factors included one or more of the following: prematurity (*n*=15, 52%), neonatal brain injury (*n*=12, 41%), and had congenital heart disease requiring neonatal surgery or prolonged hospitalization (*n*=11, 38%).

Methods: Children were assessed using the BSITD-3 (administered by a pediatric psychologist or occupational therapist) and the NSMDA at 18 to 24 months of age (corrected for prematurity). For the BSITD-3, composite motor scores of <85 and gross or fine motor scaled score of <7 are more than one standard deviation below the mean (15th centile) and were each considered delayed and appropriate for early intervention. NSMDA scores appropriate for early intervention were those classified as minimally, mildly, moderately, or severe-profoundly motor impaired.

Results: There was a significant difference in the number of children identified as delayed or impaired using the two tests (p=0.0007 by Fisher's exact test). The two assessments agreed on the classification of 23/29 (79%) infants (8 as delayed or impaired and 15 as age appropriate), but disagreed for six children who were identified as age appropriate using the BISTD-3 but impaired using the NSMDA (minimally, n=2; mildly, n=1; moderately, n=3). For these six children, further inspection of the NSMDA performance areas (gross motor, fine motor, neurologic, primitive reflexes, posture/balance, and motor responses to sensory input) revealed that 3 of the 4 infants with mild or moderately impaired development had balance and postural difficulties that are not assessed using the BSITD-3.

Conclusions: Although there was agreement between the BSITD-3 and the NSMDA for 79% of high risk children studied, the NSMDA was able to identify additional children with mild-moderate impairments of posture and balance who would benefit from early intervention services.

J4

Motor assessments in the first year of life for preterm infants predict motor outcomes at preschool age

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Background/Objectives: Preterm infants have delayed motor trajectories during the first year of life compared with infants born at term. The significance of these different patterns of development for later motor outcome is not known. This study aimed to investigate the validity of the Alberta Infant Motor Scale (AIMS) and Neurological Sensory Motor Developmental Assessment (NSMDA) during the first year of life for predicting motor development at 4 years' corrected age in very preterm children.

Design: Prospective Cohort Study.

Participants and Setting: Of 100 infants born <30 weeks' gestational age were recruited at birth from a tertiary hospital.

Methods: Infants were assessed at 4, 8 and 12 months' corrected age using the AIMS and NSMDA. The same infants were assessed at 4 years' corrected age for presence of cerebral palsy (CP) and motor outcomes using the Gross Motor Function Classification System (GMFCS) and Movement Assessment Battery for Children 2nd edition (MABC-2) by a therapist masked to previous results. Sensitivity and specificity of the AIMS (10th centile at 4 months and 5th centile at 8 and 12 months) and NSMDA (mild-profound motor dysfunction) at 4, 8 and 12 months for predicting CP and mild-severe or moderate-severe motor dysfunction (MABC-2 scores =/<15th and =/<5th centile respectively) at 4 years were calculated, along with 95% confidence intervals (CI).

Results: Of the 100 infants recruited, 87 were assessed at all 4 time points. At 4 years, 6 (7%) of the 87 children had CP (GMFCS level II=3, level III=2, level V=1), 26% had mildsevere and 12% moderate-severe motor dysfunction. The AIMS and NSMDA had 100% sensitivity (CI 46-100%) at 8 and 12 months for CP, with one CP case not identified using either measure at 4 months (sensitivity 83%, CI 36-97%). The specificity of the AIMS for CP was 83% (CI 73-90%), 79% (CI 6-87%) and 69% (CI 58-79%) at 4, 8 and 12 months respectively, whilst for the NSMDA it was 84% (CI 74-91%), 78% (CI 67-86%) and 88% (CI 78-94%). For mild-severe motor dysfunction, the AIMS had relatively low sensitivity at all time points (range 50-70%), and higher specificity at 4 and 8 months (range 88-89%) than at 12 months (75%, CI 62-85%). The NSMDA also had low sensitivity (range 35-50%), but high specificity throughout the first year (range 82-90%). For moderate-severe motor dysfunction, the AIMS had good

sensitivity at all times points (range 80–90%) and good specificity at 4 and 8 months (range 83–86%) but lower specificity at 12 months (69%, CI 57–80%). The NSMDA also had good sensitivity (range 80–90%) and specificity (range 82–88%) at 4 and 8 months, but at 12 months sensitivity was low (60%, CI 26–88%) and specificity was high (89%, CI 79–95%).

Conclusions: Motor development during the first year of life in very preterm infants assessed with the AIMS and NSMDA is predictive of later motor outcomes, although many children are falsely identified as having later dysfunction. Given that neither of these assessment tools are 100% predictive, longitudinal assessment is recommended.

J5

Developmental effects of bevacizumab treatment for retinopathy of prematurity

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Background/Objectives: Intravitreal bevacizumab (IVB) is gaining popularity as an alternative to laser therapy for infants with retinopathy of prematurity (ROP), but its systemic effects on the developing child remain largely unknown. Our goal was to determine whether infants with ROP treated with IVB have different neurodevelopmental outcomes than those treated with laser therapy.

Design: This was a retrospective cohort study of infants who received treatment for ROP with laser therapy or IVB.

Participants and Setting: All infants who received treatment for ROP at our tertiary NICU from January 2008 through September 2013 and who were seen in follow-up clinic were included. 33 infants received ROP treatment during our 57-month study period. Of those 33 patients, 2 died prior to follow-up, 6 did not keep their follow-up appointments, and 3 had appointments scheduled after the study period. 22 patients were thus included in our study. 8 of the 10 infants who received laser therapy and 10 of the 12 infants who received IVB had both eyes treated. 2 infants received laser therapy following IVB.

Methods: Clinical assessments were performed by a developmental pediatrician, neonatologist, or supervised resident. Infants were evaluated in cognitive, language, and gross motor development using the Capute and modified Peabody Developmental Motor Scales. Developmental Quotients (DQ) were calculated for prematurity-adjusted age. Parents completed Ages and Stages Questionnaires (ASQ). Infants were divided into 2 groups according to their initial treatment, laser or IVB. Means and medians for the two groups were calculated. Differences were assessed using Student's t test or Fisher's exact test, and a p value <0.05 was considered significant.

Results: Between the laser and IVB groups, the median gestational age (25w and 24.8w, p=0.97) and mean birth weight (668g and 619g, p=0.44) were not significantly different. The

incidence of grade III or IV IVH, PVL, or ventriculoperitoneal shunting were not significantly different between the two groups (p=1). The assessments occurred between 6 and 39 months, with the median follow-up age 15.2 months in both groups (p=0.5). The groups were comparable in z-scores of clinic weight, length, and head circumference. No significant differences were noted between the laser and IVB groups in global cognitive (DQ 75 and 82, p=0.47), language (DQ 73 and 93, p=0.11), or gross motor development (DQ 67 and 78, p=0.37). The groups showed similar findings in muscle tone, with 22% normal in the laser group and 33% normal in the IVB group (p=0.66). No significant difference was found in ASQ pass rate between the two groups (33% and 50%, p=0.66).

Conclusions: In patients with ROP, no significant differences in neurodevelopment were noted between those who had received treatment with IVB and those who had received laser therapy. Though our study evaluated patients with a median age older than one year, further studies are needed to assess the long-term effects of IVB on the developing child.

J6

Optimizing the motor outcomes of infants at high risk of cerebral palsy: a pilot randomized controlled trial

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Background/Objectives: Infants at the highest risk of cerebral palsy (CP) can now be identified early using the general movements assessment. Neuroplasticity and early intervention evidence favors early enrichment to optimize outcomes, yet very few trials of high risk infants have demonstrated improved motor outcomes. The aim of this study was to determine the short- term effects of 'GAME', a goal-oriented activity-based, environmental enrichment therapy programme on the motor development of infants at high risk of CP and test study procedures for a randomized controlled trial (RCT).

Design: Pilot RCT.

Participants and Setting: Infants at high risk of CP from the Sydney Children's Hospital Network, Australia.

Methods: A pilot RCT was conducted to assess motor outcomes, goal attainment, parent well-being and quality of the home environment, after 12-weeks of GAME intervention compared to standard care. GAME included: creation of movement environments to elicit new motor behaviors; parent training and support in motor learning principles and task analysis; individualised, variable and frequent functional motor task practice which emphasized self initiated movement. Data were analyzed using multiple regression.

Results: Thirteen infants at high risk of CP due to abnormal general movements during the fidgety period (3–4 months of age) were recruited and randomized to 2 groups. Infants in the GAME group (n=6) demonstrated a 5.72 point PDMS-II

advantage compared with the standard care group (n=7)(p<0.001). No significant differences existed between groups in goal attainment, parent well-being, parent satisfaction or quality of home environments after 12 weeks of intervention. Conclusions: GAME therapy appears to offer a promising new motor intervention for infants with, resulting in more favorable short-term motor outcomes than current standard practice. There is a pressing need for an adequately powered RCT with long-term end points, to determine if this type of intervention may influence the developmental trajectory of these children. The research, including recruitment and intervention was clinically feasible to do, with no study dropouts. It is timely for the field to consider how to translate neuroplasticity and motor learning evidence from older children and adults into diagnosis-specific early intervention for infants at high risk of CP.

J7

Sedentary and active time in toddlers with and without cerebral palsy

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Background/Objectives: Physical activity is important for the health and development of children. The aim of this study was to evaluate and compare levels of physical activity and sedentary behavior to physical activity recommendations for toddlers with cerebral palsy (CP) according to functional capacity (Gross Motor Function Classification System: GMFCS) and age-matched children with typical development (CTD).

Design: Cross-sectional study.

Participants and Setting: Fifty-eight children with CP (mean (SD) age 2y 6mo (6mo), 38 boys) were recruited from a population based cohort study at a tertiary care hospital. A control group of twenty children with typical development (mean (SD) age 2y 3mo (6mo), 11 boys) were recruited from the community. Children with CP were separated into three groups: GMFCS I-II (*n*=32: mean (SD) Gross Motor Function Measure: mean (SD) GMFM: 61(9%)); GMFCS III (*n*=14; mean (SD) GMFM: 44 (8%)); and GMFCS IV-V (*n*=12: mean (SD) GMFM: 30 (9%)) groups. Primary motor type was spasticity (unilateral: *n*=19; bilateral: *n*=27), dyskinesia (*n*=8) and hypotonia (*n*=4).

Methods: Children wore a triaxial ActiGraph® for three days during waking hours. Accelerometer cut-points, previously validated in our population, were applied to identify the percentage of time spent sedentary per day, minutes of active time, and the frequency and duration of sedentary and active bouts for each group. The study group validated the cut-points for CT and for children with CP according to functional capacity. A research physiotherapist performed the

GMFM-66 assessment, and classified motor type and functional severity (GMFCS I-V). ANOVA with post-hoc testing and Fisher's exact test were used to compare groups. Data are presented as mean (SD).

Results: No difference in percentage sedentary time was found between the CTD (49 (5%)) and GMFCS I-II (52 (7%)) group (Figure 1). The GMFCS III group spent more time sedentary than both CTD and GMFCS I to II groups (62 (9%), both p<0.01) (Figure 1). The GMFCS IV-V group spent more time sedentary than all other groups (74 (11%), all p<0.05) (Figure 1). Some CTD and children classified as GMFCS I to II were as sedentary as children classified as GMFCS IV-V (Figure 1). The CTD group and GMFCS I to II group were more likely to spend ≥180 minutes in active play daily on all three days than the GMFCS IV to V group (both p<0.016). The GMFCS IV to V group was more likely to have sedentary bouts ≥60 minutes than both the CTD and GMFCS I-II group (both p<0.016).

Conclusions: Differences in sedentary behavior between the CTD and mildly impaired children with CP (GMFCS I-II) were not evident in our toddler population. Children with moderate-to-severe functional impairment were progressively more sedentary and less likely to meet physical activity guidelines than CTD or children with milder impairment. Further research into the health implications of high levels of sedentary behavior in toddlers with CP is required.

J8

Indicators of readiness for independent walking in young children with cerebral palsy

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Background/Objectives: The attainment of walking is a primary goal of physical therapy intervention for young children with cerebral palsy (CP). Walking may impact independence in mobility and participation in daily activities. Knowledge of determinants of walking would guide clinical decision-making about physical therapy interventions. The objective of this study was to identify child and family factors signifying readiness for independent walking in young children with CP, Gross Motor Function Classification System (GMFCS) levels II-III. We hypothesized that child factors would have a higher association with independent walking than family factors.

Design: Secondary data analysis of an observational cohort study.

Participants and Setting: A subsample of 80 of the 429 Move & PLAY participants. Children were 2 to 6 years of age, GMFCS levels II to III and were not walking, defined as independent walking <3 steps at the beginning of the study.

Methods: The outcome variable, independent walking 3 to 10 steps identified a Walker. Six predictor variables included four child factors (functional strength, postural control, reciprocal lower limb movement, and motivation) measured one year

prior to the walking outcome, and two family factors (family support to child and support to family) reported by parents seven months after study onset. The predictive model was analyzed using backward stepwise logistic regression.

Results: After one year, 21 (26%) children were Walkers and 59 (74%) were Non-walkers. Mean scores for functional strength, postural control, and reciprocal lower limb movement were significantly higher for Walkers than for Nonwalkers, (p<0.05). Backward stepwise logistic regression showed that functional strength, operationally defined as the ability to transfer body weight during transitions into and out of standing from sitting on a bench, was the only significant predictor in the model, (odds ratio=1.45, 95% CI=1.15-1.83, p<0.05). The area under the receiver operating characteristic (ROC) curve for functional strength was (AUC)=0.71, (p<0.01), 95% CI=0.57–0.85. The best prediction of walking from the ROC curve was at a cutoff score of 3, indicating a higher probability of walking in children able to at least stand from a bench using hands. The regression model had a 79% probability of predicting not walking (negative predictive value) but only a 54% probability of predicting walking (positive predictive value).

Conclusions: A measure of functional strength and dynamic postural control in a closed chain sit to stand activity may signify readiness for independent walking in young children with CP. Further research is recommended to identify indicators of readiness for independent walking in young children with CP.

J9 Full-day monitoring to measure the quantity of kicking across the first year of life

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Background/Objectives: Very early identification of impaired infant neuromotor control is necessary for initiating and targeting early therapeutic intervention to promote optimal development. Kicking is one of the earliest motor skills infants perform, and full-day assessment is important due to high variability in infant performance. Determining the quantity, type and quality of leg movements performed by typically developing infants is necessary background data for differentiating typical, delayed and impaired developmental trajectories very early in life. The goal of this investigation is to quantify kicking rates in infants with typical development across the first year of life. We hypothesize that infant kicking rates increase across the first year of life.

Design: Prospective longitudinal cohort study.

Participants and Setting: We collected a full day (ranging from 8 to 13h) of leg movement activity from 12 infants with typical development, ages 1 to 12 months, in their homes.

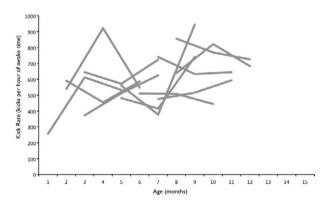
Methods: Infants were measured 3 times each, with 2 months between measurements. Gyroscope data (rate of rotation) were collected at 20 Hz from synchronized Opal sensors (APDM, Inc.) attached to the front of the ankles. We low-pass filtered the data at 4 Hz and defined a kick as a leg movement of 30 degrees/seconds of total rotation sustained for at least

0.4 seconds. We calculated descriptive statistics and used a Repeated Measures ANOVA to test for a difference in kicking rate by visit and, in follow-up, tested for a linear trend of increased kicking rate across visits using SPSS software (version 18) with an alpha level of 0.10.

Results: Overall mean kicking rate was 600 kicks per hour of awake time (SD=153), or about 10 kicks per minute when awake. Table 1 shows mean and standard deviation of kicking rate by month. Repeated Measures ANOVA did not show a significant difference in kicking rate by visit (F=1.34, df=2,22, p=0.28). Figure 1 shows the kicking rate of each infant across visits, by age. The linear trend test was significant (F=3.89, df=1,11, p=0.07).

Conclusions: These results provide guidelines for kicking rates for infants with typical development and indicate kicking rates, in general, increase across the first year of life. We will

Table 1. Kicking Rate by Age							
Age (Months)	Number Of Infants	Mean Kicking Rate Per Hour Awake	Standard Deviation				
1	1	256	NA				
2	2	565	38				
3	3	543	148				
4	2	688	330				
5	5	531	36				
6	3	548	38				
7	6	560	158				
8	3	668	176				
9	4	709	183				
10	3	678	204				
11	2	619	36				
12	2	704	30				



test for a relationship between kicking rates and developmental skill progression (for example, infants may kick less when they start sitting). We also plan to measure kicking rates in infants at risk for developmental delay. Having information about early leg movement patterns and their relationship to developmental outcomes will support early identification of typical, delayed and impaired developmental trajectories and inform early intervention practices.

Figure 1. Kicking rate by age. Each line represents an infant across 3 visits. There was a significant linear trend for increased kicking rate across visits.

J10

The effect of sensor robotic technology on the development of prone mobility in infants with or at risk of cerebral palsy

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Background/Objectives: The earliest and, in some cases, the only form of functional mobility available to infants during the first year of life is prone locomotion. Children with cerebral palsy (CP) seldom attain this important but complex milestone. The purpose of this study was to determine whether an integration of robotic and sensor technologies (SIPPC), designed for a dual purpose of influencing and measuring movement effort, would improve prone locomotion in infants with or at risk for CP. We hypothesized that that infants receiving movement assistance triggered both by the forces they apply against the floor and by specific limb movements (SIPPC-E) will demonstrate earlier and better success in achieving prone locomotion than infants receiving assistance triggered only by the forces they apply.

Design: We used a repeated measures experimental design with 2 groups: experimental group (SIPPC-E) and control group (SIPPC-C). Measures were collected twice a week for 12 weeks.

Participants and Setting: Participants were 24 infants, 4 to 6.6 months old, with or at risk for CP. Inclusion criteria: A TIMP z score = -1.0 or lower, a confirmed diagnosis of CP; or positive MRI results. The study took place in the infants' homes. Methods: The SIPPC consists of device-based wheel position sensors and limb-mounted inertial measurement units (IMUs) that measure movement performance and can be used to trigger locomotion assistance. With these sensors, measured total distance traveled and limb kinematics. Infants were randomly assigned to the experimental or control group. The training protocol consisted of three five-minute trials on the SIPPC, two times a week, for up to 12 weeks. We coded 20 combinations of arm and leg movements and used correlation and randomized two-way ANOVA with repeated measures to test the hypothesis.

Results: The mean distance for both groups increased over the

12 week period with the largest increase in the SIPPC-E group (p=0.01 for E, and p=0.067 for C). Correlation coefficients between the 20 movement patterns and the distance traveled ranged from r = 0.55-0.83 for the SIPPC-E group and r=0.32-0.56 for the SIPPC-C group. Mean excursion of arm and leg movements were higher for infants in SIPPC-E than SIPPC-C group (p-values ranged from 0.045 to 0.076). Conclusions: The differences in correlation coefficients between the experimental and control groups suggest that this approach may offer a way to bypass some of the motor control constraints that limit prone locomotion in infants with or at risk for CP. The higher arm and leg activity by infants in the experimental group also suggest that infants are capable of engaging in reinforcement learning with a complex and high dimensional movement like prone locomotion. We believe that because early prone locomotion is associated with emergence of a wide variety of skills assisting infants with or at risk

for CP, this approach will have far reaching effects. No tested interventions exist to promote the development of this skill in infants.

Free Papers K

K1

Bone mineral density in ambulatory children with cerebral palsy

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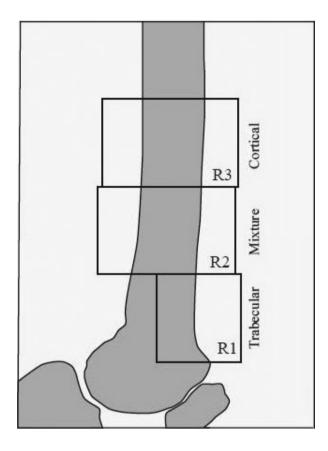
Background/Objectives: Non-ambulatory children with cerebral palsy (CP) have increased risk for low bone mineral density (BMD), mainly explained by lack of weight-bearing. However, other risk factors may be present also in ambulatory children including use of antiepileptic-drugs (AED), abnormal muscle forces on bone and poor nutritional status. The aim of the present study was therefore to assess BMD in ambulatory children with CP. We hypothesized that children with Gross Motor Function Classification System (GMFCS) level I and II would have reduced BMD, but that BMD would not differ between level I and II.

Design: Cross-sectional study.

Participants and Setting: Of 31 children (median age: 13y, 6mo; 11 girls) with CP and GMFCS level I (n=20) or II (n=11) participated in this study.

Methods: BMD (g/cm²) was assessed in the lumbar spine (LS) and in the distal femur (DF) using Dual X-ray Absorptiometry (DXA) (Hologic Discovery). In the DF, BMD was measured in three regions where region 1 comprises mainly trabecular bone and region 3 comprises mainly cortical bone, while the in-between located region 2 (R2) contains a mixture of cortical and trabecular bone (Figure 1). We used age-, sex- and race -normalized values obtained from a healthy US pediatric population to calculate z-scores. A BMD z-score of less than -2 was defined as 'low BMD' for age.

Results: We report the results obtained in the LS and in R2 of the left DF. Among all 31 participants the median BMD z-scores were -0.60 (range: -3.3; +1.3) in the LS and -1.10 (range: -5.9; +1.7) in the DF. Children with GMFCS level II had significantly lower median BMD z-score in the DF (-3.2; range: -5.9: -0.8) than children with GMFCS level I (-0.7; range: -5.5: +1.7; p<0.001). In the LS, children with GMFCS level II had a median BMD z-score of -2.0 (range: -3.3; +0.7), compared with -0.5 (range: -2.6; +1.3) in those with GMFCS level I (p=0.066). In all, 11 (35.5%) of the 31 children had low BMD for age in the DF. Of these only 2 were in GMFCS level I, while 9 were in GMFCS level II (p<0.001). In the LS 7 chil-



dren had low BMD *z*-scores, and only one of these were in GMFCS level I while 6 were in level II (*p*=0.002). Using children with GMFCS level I as reference, children with GMFCS level II had an odds ratio (OR) for low BMD for age in the DF of 40.5 (CI: 4.9–336). When adjusted for use of AED and body mass index, the OR for low BMD in the children with GMFCS level II was still increased (*p*=0.02), even if the adjusted OR decreased (OR: 21.5; CI: 1.6–288).

Conclusions: Ambulatory children with CP have reduced BMD in the LS and the DF, but in contrast to our hypothesis we found that children with GMFCS level II had significantly lower BMD in the DF than those with GMFCS level I. This may reflect differences in muscle forces across bone, despite apparent similarities in weight bearing.

K2

Influence of activity on bone health in cerebral palsy

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Background/Objectives: Low bone mass and subsequent increased risk of fractures is common in children with cerebral palsy (CP). Reduced weight-bearing is implicated in this reduced bone mineral density (BMD). The literature suggests

that ambulatory status positively influences BMD, yet the relationship between activity levels to BMD has not been examined in CP. This study examines the relationship of walking and physical activity to BMD in ambulatory children with CP. *Design:* Cross-sectional study.

Participants and Setting: Thirteen children with CP (age range 5.5–15.5y, 7 boys) were recruited through a regional pediatric tertiary care hospital. Gross Motor Function Classification System levels were represented by 9, 2 and 2 participants for levels II through IV respectively. Pubertal staging was age appropriate for all but one patient whose pubertal staging was advanced for age based on parental report using a validated self/family-pubertal stage reporting tool.

Methods: All participants under went Densitometry (DXA) of the lumbar spine and lateral distal femur (3 regions) and vitamin D (25-OH total) levels were obtained. Walking activity was captured with the variables of average total strides/day, percent of active time walking and average number of strides/day at >30 strides/min via a 5 day sample with a StepWatch accelerometer on the left ankle. Physical activity was documented through parental report of the Activities Scale for Kids (ASKp-30) summary score. Relationships were examined through Spearman rho correlations.

Results: The BMD z scores for the lumbar spine and femur ranged from -3.1 to 0.10 and -4.2 to 1.0 respectively. Vitamin D levels averaged 26.08 [11, 35]. Average total strides/day [range] were documented at 4554 [250, 9760] with percent time walking 0.42 [0.10, 0.70] and average number of strides >30 strides/minutes 1906 [13, 5142]. ASKp- 30 scores averaged 64.0 [19, 87]. Walking and physical activity was not significantly related to lumbar spine z- scores. Average total strides/day (0.67, p=0.02) and average number strides >30 stride/minutes (0.67, p=0.02) correlated with femur region 1 z-scores but not femur regions 2 and 3 (0.23 to 0.56, p=0.06 to 0.47). ASKp-30 correlated with all three femur regions z-scores (0.70 to 0.83, p=.01). Vitamin D levels did not correlate with DXA scores.

Conclusions: While BMD is frequently not tested in ambulatory youth with CP, this work suggests that they are at risk for poor bone mineral density and subsequent bone health issues. Results also suggest that accelerometry-based walking activity levels and intensity appears related to higher BMD in the cancellous bone of the distal femur (region 1). Parent also reported physical activity is positively related to BMD in all regions of the distal femur. Further work should examine the influence of habitual walking levels and intensity on BMD with potential implications for clinical monitoring and/or interventions for ambulatory youth with CP.

K3

Heart rate variability in children with cerebral palsy and acquired brain injury

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Background/Objectives: Children with cerebral palsy (CP) may demonstrate cardiac autonomic function (CAF) disturbance. As CAF may be associated with motor function it is important to assess this association in children with CP in comparison to children with acquired brain injury (ABI) and typically-developed children (TDC). A non-invasive measurement of CAF is the heart rate variability (HRV). Since CAF may predict clinical outcomes it is important to evaluate its' component with various activities. *Objectives*: (1) to evaluate CAF during rest and exercise in children with CP, ABI and TDC; and (2) to assess the association of CAF with mobility.

Design: Cohort retrospective study.

Participants and Setting: Nineteen children with CP, eleven children with ABI, and eight TDC participated. The evaluation took place in a rehabilitation out-patient clinic as part of their yearly follow-up.

Methods: CAF was established via heart rate variability (HRV) with a Polar watch and assessed via the following measures: mean R-R intervals, standard deviation of the average R-R intervals, root mean square-standard deviation and high and low frequencies. HRV was assessed during rest, aerobic and isometric exercise. Isometric exercise (children with CP only) consisted of pushup with a graviton machine. Aerobic exercise consisted of the six minutes walk test (6MWT). Mobility level was established via the distance walked in the 6MWT and gait speed in 10 meters. One way ANOVA examined differences between the groups in HRV. t-test compared rest and exercise HRV. Associations were assessed via Pearson correlations.

Results: Compares to TDC, at rest, children with CP and ABI demonstrated lower HRV. Several HRV measures at rest of children with CP were lower than these of children with ABI. HRV changed significantly during aerobic exercise only among the TDC. Similarly, HRV indices of children with CP during isometric exercise and rest were not significantly different. The decrease in HRV in children with CP and ABI were related to mobility level. In conclusion, compares to TDC, the cardiac autonomic mechanism of children with CP and ABI is less efficient at rest, less adaptive to exercise and related to mobility. However, children with CP exhibited greater cardiac autonomic mechanism dysfunction.

Conclusions: Compares to TDC, the cardiac autonomic mechanism of children with CP is less efficient at rest, less adaptive to exercise. Inspecting the different efforts performed (aerobic and isometric), the CP children show little adaptation to various type of exercise. Therefore, children with CP exhibited greater cardiac autonomic mechanism dysfunction. Significance: The autonomic nerve system reaction to exercise of Children with CP and BI is different.

K4

The role of bladder management and functional mobility in promoting life satisfaction over time for individuals with childhood-onset disabilities

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Background/Objectives: Urinary incontinence is a common problem for individuals with physical disability. Previous

research highlights the role of proper bladder management in optimizing psychosocial functioning and preventing urological complications. The objective of the current study was to build on this research by exploring the interrelationship of bladder management and functional mobility in predicting life satisfaction across the lifespan for individuals with childhood-onset spinal cord injury (SCI).

Design: Longitudinal follow-up study.

Participants and Setting: A total of 356 adults (ages 19–39, M=27.17, SD=3.64) who were current or former patients from one of three pediatric SCI programs in the United States were recruited for this study. All participants sustained a SCI prior to age 19 (M=14.32, SD=4.47) and had been injured an average of 12 years (SD=8.76, range 4–30). 64% were male, 86% Caucasian, 43% had paraplegia, and 70% complete injuries.

Methods: Participants completed measures assessing demographic and injury characteristics, life satisfaction (SWL), and functional independence (FIMTM) on approximately an annual basis. A total of 1858 interviews were conducted, and a majority of participants contributed to at least 3 waves of data (65%; Range 2–14; *M*=5.34; SD=2.99).

Results: A multilevel modeling approach was used to investigate the relationship between bladder program and functional motor abilities on life satisfaction. The motor subscale of the FIM was used to characterize impairment and disability in motor functions. Results suggest, that for individuals with urinary incontinence, both intermittent catheterization and continent diversion bladder programs are associated with significantly higher life satisfaction scores than other treatment approaches (β =2.56, SE=1.24, p=0.04). Additionally, functional motor skills moderated this effect such that the association between type of bladder management and life satisfaction is greatest when functional mobility is also high (β =0.16, SE=0.04, p<0.01). Thus, being higher in overall mobility appears to amplify the importance of appropriate bladder program on life satisfaction.

Conclusions: These findings underscore the importance of bladder management programs for life satisfaction, especially for those with greater mobility.

Kı

A population-wide intervention to improve the care of children with medical complexity

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Background/Objectives: Children With Medical Complexity (CMC) experience frequent interactions with the medical system. Despite the benefit of modern technology, CMC often receive care that is costly, duplicative, and inefficient. CMC comprise 6% of children on Medicaid, yet consume 40% of Medicaid dollars spent on children. We created a population-wide initiative to improve the care of CMC. Our goals comprised a 'triple aim': to reduce the cost of care, improve the

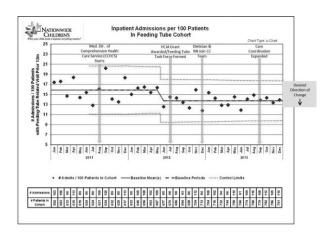


Figure 1: Reduction in Inpatient Admissions

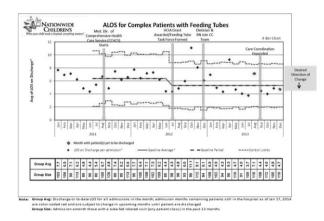


Figure 2: Reduction in Average Length of Stay

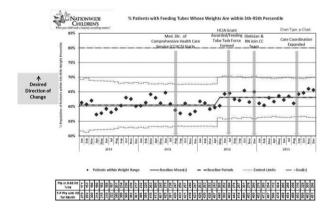


Figure 3: Improvement in Weight Status

quality of care, and improve the nutritional status for CMC in our 34 county catchment area.

Design: Population-based cohort study.

Participants and Setting: Children aged 0 to 18 years with a percutaneous feeding tube and a neurologic diagnosis seen at Nationwide Children's Hospital within the past 12 months with Medicaid as their primary payor.

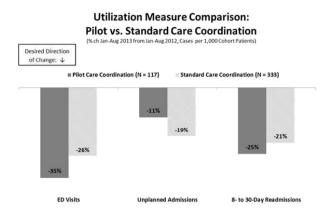


Figure 4: Improvements attributable to enhanced care coordination

Methods: System-wide, interdisciplinary subcommittees examined difficulties in our system, created interventions, and developed key driver diagrams focused on our triple aim goals. Key interventions included: (1) Electronic Medical Record Standardization. (2) Standardized Feeding Tube Education for Families (3) Standardized care of feeding tubes (4) Standardized involvement of Dietitians (5) Proactive Care Coordination that meets National standards The 2 sample Poisson test was used to compare groups as appropriate.

Results: There are approximately 615 children in our cohort; this number changes monthly. (1) Cost reduction: There was a 17.5% decrease in the average length of stay (Baseline 12mo average=6.4d; Current 12mo average=5.2d) and an 11.2% decrease in the number of admissions (Baseline 12mo average=5.6 admissions/100 cohort patients; Current 12mo average=13.8 admissions/100 cohort patients). Both of these decreases resulted in a baseline shift. (2) Improved Care: We engaged 117 children in a pilot of proactive care coordination (26% of the cohort-limited to those with a tube in 2012 and 2013). In this group, there was a 35% decrease in ED visits, an 11% decrease in unplanned admissions and a 25% decrease in readmissions compared to the same period in the previous year. The other children in the cohort experienced a 26% decrease in ED visits, a 19% decrease in unplanned admissions, and a 21% decrease in readmissions over the same period. While differences between groups were not statistically significant, this pilot helped us establish and implement our institutional standard for care coordination. (3) Improved Health: We observed a 5% increase in the percent of tube fed children with weights between the 5th and 95th centile on a standard growth curve (Baseline 12mo average=60%; Current 12mo average=63.1%). Conclusions: Our multipronged approach to population health has shown promising preliminary results including reduced cost for the care of CMC while improving their health and quality of care. Vital elements of the process include standardizing care across multiple providers and venues, engaging in proactive care coordination, and improving education of families and caregivers.

K6

Is growth and nutritional status in children with cerebral palsy related to the severity of the brain lesion?

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Background/Objectives: This study aimed to investigate the relationship between brain lesion severity assessed on structural magnetic resonance imaging (MRI) and growth and nutritional status in preschool-aged children with cerebral palsy (CP).

Design: Prospective population based cohort study.

Participants and Setting: Of 103 children with CP (61 male, 56 born at term, 19 preterm and 28 very preterm), with mean (SD) age 2 years 11 month (6mo), were recruited from the community. GMFCS levels were: I=51, II=5, III=17, IV=11, V=19. Motor type was bilateral spasticity (n=55), unilateral spasticity (n=30), and 18 other motor types. MRI scans were classified using Kragheloh Mann (KM) criteria into 5 groups: brain maldevelopment (BM) (n=5), periventricular white matter lesions (PWM) (n=58), cortical and deep grey matter (CDGM) (n=23), miscellaneous (M) (n=9) or normal (n=8). Methods: Brain lesion severity was scored by a trained child

neurologist using a semi-quantitative scale (possible range 0 to 40). Height (n=40) or length (n=47) were measured to the last completed millimetre directly or estimated from knee height (n=16). Weight was measured to the nearest 100 grams and body mass index (BMI) was determined. Anthropometric data were converted to Z-scores to adjust for age and sex. Results are mean (SD). Data analysis was by ANOVA and post hoc independent t-tests, Pearson's correlation coefficients and linear regression models (SPSS). Significance was set at p<0.05. Results: Children with GMFCS III–V compared to GMFCS I and II, were shorter (height Z-score: -0.14 (1.19) vs -0.85

(1.23)), lighter (weight Z-score: -0.03 (1.11) vs -1.02 (1.61))

and had lower BMI's (BMI Z-scores: 0.13 (1.23) vs -0.51 (1.64)). No difference was found in any of the anthropometric variables according to qualitative MRI classification (KM). Height and weight Z-scores were related to global MRI severity score (r=-0.51 and 0.36), summary hemispheric scores for both sides of the brain (r=-0.44 and -0.33) and summary basal ganglia and brain stem scores (r=-0.44 and -0.31) for the children with PWM injury only. BMI Z-scores were not significantly related to lesion severity scores for any group. When preterm status was taken into account, the relationship remained significant for height and weight Z-scores and global MRI severity score (β =-0.49 and -0.34), summary hemisphere scores (β =-0.41 and -0.29), and summary basal ganglia and brain stem scores (β =-0.42 and -0.30).

Conclusions: Brain lesion severity in children with PWM was related to height and body weight even after preterm status was considered. Children with more severe PVM were shorter and lighter than those with less severe lesions. These results

suggest the need for more accurate nutritional care in patients with more severe brain lesions in the PWM group. Further investigation into the ability of this semi-quantitative MRI scale to predict future growth and nutrition outcomes is warranted.

K7

Energy and micronutrient intakes of pre-school aged children with cerebral palsy

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Background/Objectives: This study investigated micronutrient intakes and their association with energy intake in pre-school aged children with cerebral palsy (CP).

Design: Prospective population based cohort study.

Participants and Setting: Of 106 children with CP (71 male), mean (SD) age 2 years 7 month (10mo) were recruited from the community. GMFCS levels: I=51, II=15, III=15, IV=8, V=17. Primary motor type was bilateral spasticity (n=54), unilateral spasticity (n=37) and 15 other motor types. 94 children were orally fed and 12 children were tube fed.

Methods: Energy and micronutrient intake was determined using a parent completed validated three-day weighed food record, and was analysed using the Foodworks TM dietary analysis software (2009; Xyris Software). Height or length were measured to the last completed millimetre or estimated from knee height. Weight was measured to the nearest 100 grams. Anthropometric data were converted to Z-scores. Data are presented as mean (SD). Significance was set at p<0.05. Data analysis utilized ANOVA and Pearson's correlation co-efficients and partial correlations (SPSS).

Results: Height Z-scores were significantly lower for children classified as GMFCS II (-1.0 (1.6), III (0.9 (1.5)), and IV (1.4 (1.6)) than those with GMFCS levels I (-0.1 (1.0)). There was no significant difference between GMFCS I and V (-0.7 (1.4)). There were no significance differences in weight or BMI Z-scores between groups. Children classified GMFCS V, had significantly lower energy intakes (3.1 (0.8)MJ/day), compared with GMFCS I (4.5 (1.0)MJ/day), II (4.2 (0.9)MJ/day), III (4.1 (1.5)MJ/day) and IV (4.3 (1.2)MJ/day). Daily nutrient intakes for all children were: calcium 670 (313)mg, iron 7.5 (9.9)mg, zinc 5.6 (2.6)mg and vitamin C 62.8 (66.6)mg. Children with intakes less than the recommended dietary intake (RDI) for each nutrient were: iron n=85 (80%), calcium n=36(34%), vitamin C n=36 (34%) and zinc and n=10 (9%). Those with intakes less than 80% of the RDIs were: iron n=77(73%), calcium n=25 (24%), vitamin C n=31 (29%) and zinc n=4 (4%). Children with greater calcium intakes had significantly lower intakes of iron (r=0.19), even when tube feeding was controlled for (r=0.20). Children with greater energy intakes had significantly greater intakes of calcium (r=0.47), total-folate (r=0.43), sodium (r=0.63), potassium (r=0.40), magnesium (r=0.70), phosphorus (r=0.73), zinc (r=0.73) and iodine (r=0.37). These relationships remained significant when tube feeding was taken into account. There were no significant associations between energy intake and iron, vitamin C, thiamin, riboflavin, and vitamin E intake.

Conclusions: These results show many preschool aged children with CP are not meeting RDIs for several micronutrients. Only certain key micronutrients are related to energy intake and as children consume more calcium containing foods they consume less iron. To optimize the nutritional status of children with CP micronutrient intake needs to be considered in addition to energy intake.

K8

Assessment of the impact of an enteral nutrition method (bolus vs continuous infusion) on carbohydrate metabolism in children with neurological impairment fed by percutanous endoscopic gastrostomy

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Background/Objectives: Continuous enteral nutrition (EN) given by PEG (percutaneous endoscopic gastrostomy) is often the only possible way to obtain adequate body mass and length increases in malnourished children with neurological impairment not tolerating EN provided in boluses. That manner of feeding, as non physiological one may potentially induce carbohydrate metabolism disorders and entail risk of vascular complications. Literature offers studies that only assess carbohydrate metabolism in enterally fed patients with diabetes mellitus; there are no data concerning other groups of patients, especially children with neurological impairment receiving long-term EN. Design: Retrospective cohort study.

Participants and Setting: Of 39 patients (24 girls, 15 boys, mean age 8.5 [5.38] years) with neurological impairment receiving Home Enteral Nutrition Service for at least 6 months were enrolled to the study.

Methods: 25/39 of those were fed by boluses (4-6 a day, Bf group), while 14/39 received continuous infusion (min/max 18-20h, Cf group). Short and long-term indices of glucose control expressed by blood glucose concentration and glycated hemoglobin (HbA1C) were measured in blood samples. Blood glucose level was tested in the morning at least 4 hours. After feeding. According to International Diabetes Federation level 70 to 99 mg/dl was considered normal glucose concentration and 4.8 to 6.0% was the normal range of HbA1C.

Results: Mean glucose concentration in both the group was 83.69 (8.5) mg/dl (Bf: 81.56 [4.25]mg/dl; Cf: 87.48 [6.5]mg/ dl; NS). Frequency of hyperglycemia was also comparable (3/ 25 in Bf vs 2/14 in Cf, NS). On the other hand, hypoglycemia was more common in children fed by boluses compared to children nourished by continuous infusion (6/25 vs 1/14 chi² test p=0.046). Mean HbA1C level in the whole study group was 5.04 (0.43)% No differences in HbA1C depending on the feeding method were found (mean HbA1C in group Bf was 5.00 [0.36] vs 5.11 [0.53]% in group Cf, NS). Also, there were no differences in the incidence of abnormal HbA1C results in both the groups (Bf: 1/14 vs Cf: 0/25; NS and Bf: 7/25 vs Cf: 3/14; NS). Mean diet energy value for children fed by boluses was lower than for children fed by continuous infusion (62.3kcal/kg in group Bf vs 73.36kcal/kg in group Cf).

Conclusions: (1) Continuous enteral nutrition does not enhanced the risk of short and long-term hyperglycemia in children with neurological impairment. (2) However, bolus enteral feeding may be associated with the risk of hypoglycemia in this group of patients.

K9

Adipose tissue distribution and musculoskeletal density in adults with cerebral palsy

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Background/Objectives: Premature declines in function among adults with cerebral palsy (CP) may occur as a result of early and accelerated atrophy and weakness, beyond that which is expected for typically-developed adults. Despite evidence to confirm that individuals with CP have less lean body mass and greater relative adiposity, the interrelationships between altered fat partitioning and muscle and bone quality is less well-understood. The purposes of this study were therefore to examine the differences in trunk adipose tissue distribution, lumbar bone mineral density (BMD), and muscle attenuation (i.e., an indicator of intermuscular adiposity) in adults with and without CP, and to determine the association between adipose tissue, muscle attenuation, and BMD.

Design: Cross-sectional.

Participants and Setting: Participants were from a convenience sample of 41 adults with CP (GMFCS I-V; age: 38.8 [14.4] years; weight: 61.3 [17.1]kg), with existing thoracic and abdominal computed tomography (CT) scans, and a cohort of 311 typically-developed and available clinical patients, matched for sex, age, and body mass. Of the 41 participants with CP, 10 participants were not match-able due to very low body mass, and thus were included as a comparison subset. Methods: Trunk fat distribution (i.e., visceral and subcutaneous adipose tissue areas), psoas major area and attenuation in Hounsfield units (HU), and cortical and trabecular BMDs in

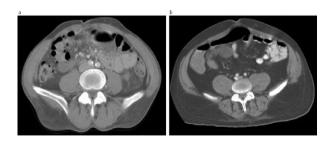


Figure 1: Computed tomography image at vertebral level L4, depicting trunk adiposity distribution and muscle size in: (a) a 53 year old, typically-developed male (65kg body mass), and (b) a 54 year old male with CP (66kg body mass).

HU were determined from existing CT scans. All morphologic assessments and comparisons were determined from the L4 vertebral level.

Results: Typically-developed adults had significantly higher cortical (β =63.41, p<0.001) and trabecular (β =42.24, p<0.001) BMD, as well as psoas major area (β =374.51, p<0.001) and attenuation (β =9.21, p<0.001), as compared to adults with CP, even after adjustment for age, sex, and body mass. Conversely, adults with CP had significantly greater adiposity in both the visceral (β =3914.81, p<0.001) and subcutaneous (β =4615.68, p<0.001) depots. Among the adults with CP, muscle attenuation was significantly correlated with trabecular (r=0.51, p=0.002) and cortical (r=0.46, p=0.006) BMD; whereas, visceral adipose tissue was negatively associated to cortical BMD $(\beta = -0.037 \text{ cm}^2; r^2 = 0.13; p = 0.03)$. Compared to the 31 matched participants with CP, the unmatched comparison-subset of adults with CP had significantly lower body mass (45.49 [8.33]kg vs 66.34 [16.05]kg; p<0.001) and psoas major area $(1359.58 [600.31] \text{mm}^2 \text{ vs } 2049.51 [1074.84] \text{mm}^2; p=0.03), \text{ but}$ less visceral adiposity (5035.45 [2888.77]mm² vs 10047.45 [8536.60]mm²; p=0.01) and subcutaneous adiposity (12796.58) [7639.65]mm² vs 21344.50 [14929.41]mm² p=0.04).

Conclusions: Adults with CP have significantly lower cortical and trabecular BMDs, smaller psoas major area, greater intermuscular adipose tissue, and greater visceral and subcutaneous adiposity than matched, typically-developed adults. Among adults with CP, muscle attenuation and BMD was positively correlated, and visceral adiposity and cortical BMD were inversely associated.

K10 Down syndrome growth charts and trends in obesity

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Background/Objectives: It is well known that children with Down syndrome (DS) tend to have shorter stature and are at risk for obesity. Large sample growth charts for such children in the U.S. do not appear to be available.

Design: Observational longitudinal cohort study.

Participants and Setting: We identified persons age less than 25 with DS from the California Department of Developmental Services between 1985 and 2010. We abstracted height and weight measures, along with information on comorbidities and functional disabilities, from annual Client Development Evaluation Reports. We excluded children with cancer, degenerative disorders, circulatory disease, or congenital anomalies (not including congenital heart defects), traumatic brain or other acquired injuries, autism, cerebral palsy, or chromosomal anomalies other than DS.

Methods: We estimated growth centiles for ages 2 to 20 using generalized additive models and compared them with the U.S. general population (GP). We used generalized estimating equations to determine whether height, weight, and BMI varied with sex, congenital heart defects, epilepsy, intellectual disability, ambulation ability, feeding ability, or calendar year, all after controlling for age.

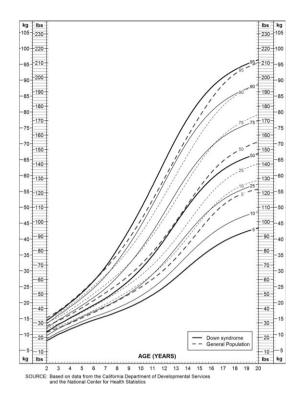


Figure 1: Weight centiles for boys aged 2 to 20 with DS and in the GP.

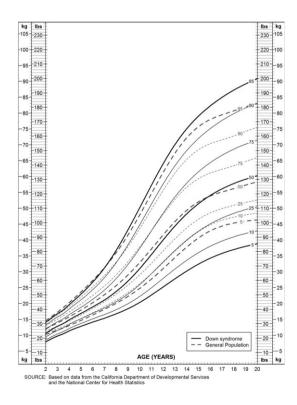


Figure 2: Weight centiles for girls aged 2 to 20 with DS and in the GP.

Results: We obtained 74 398 height and weight measurements from 17 910 children (56% male) with DS. Most were ambulatory (88%), fed themselves (85%), and did not have severe or profound intellectual disability (89%). Fewer than 1% had epilepsy, and 14% had congenital heart defects. In adolescence and early adulthood, the heaviest DS children were heavier than their counterparts in the GP, while conversely the lightest DS children were lighter (Figures 1-2). As in the GP, DS boys were heavier and taller than girls. On average, children with DS were noticeably shorter and had higher BMI than in the GP. Height and weight were lower in children with severe or profound intellectual disability (p<0.001), epilepsy (p<0.001), and congenital heart defects (p<0.001). Children older than 5 years who did not walk or self-feed also had lower height (p<0.01) and weight (p<0.001). BMI steadily increased from 1985 to 2000 (p<0.001), yet plateaued from 2000 to 2010. In the late 1980s, 20% of children aged 12 to 19 met the criteria for childhood obesity. This proportion increased to 40% in the year 2000 and remained fairly con-

Conclusions: We constructed growth charts using the largest known sample of children with DS. As expected, children with DS were shorter and had higher BMI than those in the GP. Children with comorbidities had significantly lower height and weight. Although obesity rates increased from the mid-1980s to the year 2000, they appear to have stabilized since.

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L1

Neonatal physiological correlates of early brain development on MRI and DTI in very-low-birthweight preterm infants

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Background/Objectives: Structural brain abnormalities at nearterm age have been recognized as potential predictors of neurodevelopment in children born preterm. The aim of this study was to examine the relationship between early brain development and physiological risk factors in very-low-birthweight (VLBW) preterm infants at near-term age, based on structural brain MRI and semi-automated, atlas-based diffusion tensor imaging (DTI) analysis.

Design: This ongoing prospective cohort study examines relations between neonatal physiological risk factors and white matter (WM) microstructure on DTI at near-term age.

Participants and Setting: A cross-sectional sample of 102 VLBW preterm infants (BW≤1500g, GA≤32wks) were admitted to the Lucile Packard Children's Hospital NICU and

recruited to participate prior to standard-of-care MRI conducted at 36.6 (1.8) weeks postmenstrual age from 2010 to 2011; 66/102 also received DTI.

Methods: Brain abnormalities were assessed on structural MRI, and WM microstructure was analyzed in six subcortical regions defined by DiffeoMap neonatal brain atlas. Regions of interest included the genu and splenium of the corpus callosum, anterior and posterior limbs of the internal capsule, the thalamus, and the globus pallidus, using a threshold value of fractional anisotropy (FA)>0.15 to select WM. Regional FA and mean diffusivity (MD) were calculated and examined in relation to early physiological risk factors including: bronchopulmonary dysplasia (BPD), sepsis, necrotizing enterocolitis (NEC) and level of thyroid stimulating hormone (TSH) during the hospital stay, as well as serum glucose, albumin, total bilirubin, and C-reactive protein (CRP) levels during the first two weeks of life.

Results: Of 38/102 infants had brain abnormalities observed on MRI. These infants had significantly higher mean serum CRP levels (0.64 vs 0.34 mg/dl, p=0.008) and peak CRP levels (1.57 vs 0.67mg/dl, p=0.006) compared to those with no observed brain abnormalities. Number of signal abnormalities observed on MRI correlated to serum CRP levels (r=0.302, p=0.003). Mean genu MD was higher in males than females (p=0.004). Higher thalamus MD on the left and right, respectively, was associated with lower GA-at-birth (r=-0.322, p=0.009; r=-0.381, p=0.002), lower BW (r=-0.287, p=0.021; r=-281, p=0.024). Higher thalamus MD on the left and right was also associated with lower mean serum albumin (r=-0.276 p=0.029; r=-0.385, p=0.002) and with lower total bilirubin (r=-0.293, p=0.020; r=-0.337 p=0.007).

Conclusions: At near-term age, the thalamus WM microstructure may be particularly vulnerable to neonatal risk factors. Interactions between albumin, bilirubin, phototherapy, and brain development warrant further investigation. Identification of physiological risk factors associated with selective vulnerability of certain brain regions at near-term age may clarify the etiology of neurodevelopmental impairment and inform neuroprotective treatment for neonates.

L2

Relationship between white matter fractional anisotrophy and general movement assessments in high-risk preterm infants: a tract-based spatial statistical analysis

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Background/Objectives: The quality of infant general movements, specifically fidgety movements (FMs), may be a marker of early brain impairment and may reflect the integrity of white matter (WM). Tract-based spatial statistics (TBSS) is a new observer-independent tool that analyzes multi-subject dif-

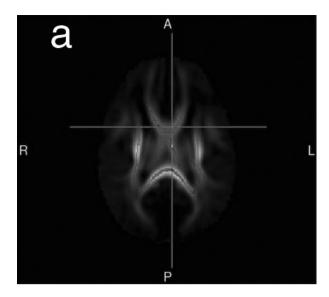


Figure 1: Voxels showing a significant correlation between FA and FM are in red-orange.

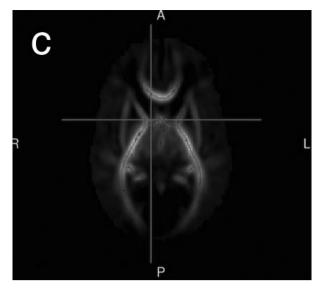


Figure 3: Voxels showing a significant correlation between FA and FM are in blue.

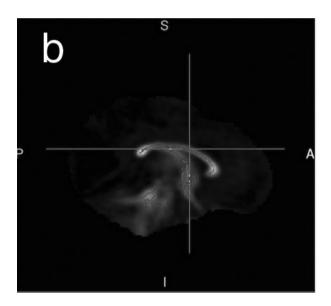


Figure 2: Voxels showing a significant correlation between FA and FM are in red-orange.

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Figure 4: Voxels showing a significant correlation between FA and FM are in yellow.

fusion data and WM microstructure. Our objective was to use TBSS to describe the relationship between WM microstructure at term age equivalent (TAE) in preterm infants and general movement assessment (GMA) outcome at 10 to 15 weeks post-term age (PTA).

Design: Prospective cohort study design of very preterm children enrolled in a neuroprotective clinical trial.

Participants and Setting: Of 45 infants born at ≤31 weeks gestational age, with a birth weight of less than 1500 grams, who required oxygen at birth, were recruited at a university hospital. *Methods*: All infants received a cerebral MRI scan on a 3T Phillips Achievea MRI using a SENSE MRI coil array, at

TAE. The GMA was performed at 10 to 15 weeks PTA with video analysis by two expert GMA certified testers using Prechtl's methodology. FMs were categorized as absent, sporadic, intermittent, or continuous based on their temporal organization (i.e. their occurrence during the video recording). Fractional anisotrophy (FA) coefficients were calculated by using the Diffusion Toolbox in the FSL package. All participants' FA images were aligned to a target in a common space by using an optimized TBSS protocol for neonates. The aligned images were used to create a mean FA map and a mean FA skeleton, which represented the centers of all tracts common to the group. Voxelwise cross-subject statistics was performed to assess the relationship between FA and GMA. In all analyses, a p value <0.05 was considered statistically significant.

Results: Of 28 infants had intermittent or continuous FMs, 10 had absent FMs, 6 had sporadic FMs and 1 was excluded because of gross pathology. The group of infants with present FMs (sporadic, intermittent, or continuous) had significantly larger FA coefficients in the corpus callosum genu, body, and splenium (Figure A), and in the fibria fornix (Figure B). The group of infants with absent FMs had significantly larger FA coefficients in the anterior limb of the internal capsule, right and left (Figure C). When examining the temporal organization of FMs from absent to continuous, FA was significant in the corpus callosum forceps major and a small portion of the posterior limb of the internal capsule (Figure D).

Conclusions: An increase in FA in specific regions of WM in preterm infants at TAE was related to the presence or absence of FMs and their temporal organization. These findings suggest FMs may be an indicator of structural connectivity and a biomarker for future neurodevelopmental activity outcomes.

L3

Neuroplastic sensorimotor resting state network reorganization in children with hemiplegic cerebral palsy treated with constraint-induced movement therapy

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Background/Objectives: A large volume of clinical evidence suggests that constraint-induced movement therapy (CIMT) effectively improves functionality by addressing learned nonuse for individuals with hemiplegic cerebral palsy (CP). Motor task-based fMRI has variably revealed possible neural reorganization following CIMT, however resting state fMRI (RSfMRI) in the absence of task has many advantages in acquisition, analysis, and interpretation. The primary objective of this study was to evaluate any clinically correlated resting state network reorganization in hemiplegic CP participants following CIMT.

Design: Prospective case-comparison cohort.

Participants and Setting: Of 11 children with hemiplegic CP (age range 6–17y, 6 cases) were recruited from Holland Bloorview Kids Rehabilitation Hospital, Thames Valley Children's Treatment Centre and McMaster Children's Hospital. Comparison participants were recruited prospectively based on the CIMT-treated group's baseline Quality of Upper Extremity Skills Test (QUEST) scores (±15).

Methods: Individuals were evaluated clinically and participated in an MRI session at baseline, 1-month and 6-months later (control participants were only evaluated at two time points) using the QUEST, Jebsen-Taylor Tests of Hand Function and Canadian Occupational Performance Measure (COPM). The sensorimotor resting state network was found using ICA

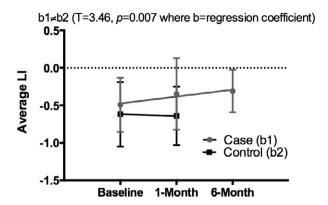


Figure 1.

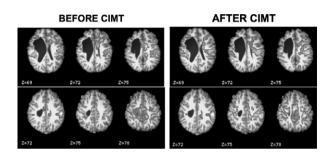


Figure 2.

and dual regression algorithms and from this laterality indices (LI) were quantified between ipsilesional and contralesional motor areas. The changes in clinical and neuroimaging measures reflect baseline scores subtracted from 1-month or 6-month scores. Paired-sample *t*-tests evaluated the change in RSfMRI LI between and within groups. Correlational analysis explored relationships between change in neural and clinical outcomes in the case group.

Results: Damage in ipsilesional motor areas often caused unilateral baseline sensorimotor network organization. The LI at 6-months approached 0 (i.e. symmetric) and was significantly different from baseline (p=0.034). The LI based on the number of voxels did not change significantly in the control (p=0.70) or case group (p=0.070) at 1-month, however the slopes describing the overall change were significantly different between groups (Figure 1). In the case group, the correlation between the change in QUEST scores at 1-month and the change in LI based on the number of above-threshold voxels was not significant (r=0.79, p=0.062). There was a significant correlation between the difference in COPM scores at 6-months and the change in LI (r=0.82, p=0.046).

Conclusions: The resting state sensorimotor network reorganizes into a more bilateral connectivity pattern with increased involvement from ipsilesional motor areas after CIMT (Figure 2). This reorganization persisted 6-months later. Spatial and quantitative resting state changes correlated with clinical changes after therapy, though power was limited by small sample size. This result is congruent with previous task-based fMRI studies and provides further evidence of the neuroplastic response to CIMT.

L4

Using diffusion tensor imaging as a surrogate to identify corticospinal tract connectivity in children with unilateral spastic cerebral palsy

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Background/Objectives: (A) to determine if Diffusion Tensor imaging (DTI) can be a reliable surrogate for identifying Corticospinal Tract (CST) connectivity in children with unilateral spastic cerebral palsy (USCP), and (B) to determine if there are structural differences between the more-affected versus the less-affected CST as assessed by DTI in children whose CSTs originate in both hemispheres of motor cortex. We hypothesized that: (a) DTI can be a sensitive and specific surrogate of transcranial magnetic stimulation (TMS), a validated physiological measure of CST connectivity, for identifying CST connectivity in children with USCP, and (b) that the integrity of the more-affected CST would be worse than the integrity of the less-affected CST, as measured by DTI. Design: Prospective cohort study.

Participants and Setting: Twenty children with USCP participated in this study (ages between 6–17y, mean age=9 years old; 16 boys, 4 girls; 10 right-sided hemiplegia, 10 left-sided hemiplegia; MACS: level I (*n*=3), level II (*n*=14), level III (*n*=3)); university laboratory.

Methods: We mapped the cortical representation of the more-affected hand using single-pulse TMS. We used the TMS-derived motor map as a seed (region of interest, ROI) to visualize the CST in DTI tractography off-line. We verified the presence or absence of the CST controlling the more-affected hand projecting from each hemisphere by comparing TMS maps and DTI tractography. We used Fisher's exact test to determine the consistency between TMS and DTI. We calculated the sensitivity and the specificity of DTI determining CST connectivity, using TMS as the 'gold standard'. Finally, we used paired t-test to compare the values of fractional anisotropy (FA) between the more-affected and less-affected CSTs in children with bilateral connectivity.

Results: There was a high correlation between TMS and DTI in determining the origins of CST controlling the more-affected hand (Fisher's exact test, p=0.0001). The sensitivity of DTI for determining CST connectivity was 93.1%, and the specificity was 77.8%. Nine children had CSTs descending from both motor cortices (i.e., bilateral connectivity) as determined by DTI. FA value of their less-affected CST was significantly larger (indicating better structural integrity) than their more-affected CST (FA of the less-affected CST=0.533, FA of the more-affected CST=0.495; paired t-test, p=0.003).

Conclusions: CST connectivity assessed by DTI replicates findings with TMS mapping. Such maps have been used to guide rehabilitation therapies. Therefore, DTI-identified CST connectivity may also be valuable for assigning intensive hand therapy and for identifying targets for brain stimulation, as TMS excludes children with seizure history and it may not be accessible to children. This study provides neuroimaging evidence that the more-affected CST is structurally compromised compared to the less-affected CST in children with bilateral connectivity.

L5

Relationship between brain lesion severity and ambulation in preschool aged children with cerebral palsy

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Background/Objectives: To assess the relationship between brain lesion severity (structural MRI), ambulatory status and gait patterns in pre-school aged children with cerebral palsy (CP). Design: Cross sectional prospective population based cohort study.

Participants and Setting: Of 150 children (mean age 3.8,

SD=1.8y, n=86 males including GMFCS level I=60 (40%), II=22 (15%), III=24 (16%), IV=22 (15%), V=22 (15%); diagnosed with CP who attended tertiary and regional referral centres were assessed on motor capacity, ambulatory status and gait patterns between 24 and 60 months corrected age. Methods: Structural MRI neuroimaging (T1, T2, Flair) performed at mean age 1.9 (SD=1.4) years were classified with (1) novel semi-quantitative scale for structural MRI (global severity score from 0 to 40, Fiori, 2013) and (2) Krageloh-Mann's qualitative classification of brain lesion pattern according to location, extent and presumed timing. Children's ambulatory status (ambulant unaided, ambulant aided, non-ambulant) and gait pattern classification for unilateral CP (Winter, Gage & Hicks, 1987) and bilateral spastic type CP (Sutherland & Davids, 1993; Rodda, 2004) were assessed by two experienced physiotherapists. The association between MRI global severity scores and qualitative MRI classification was compared to ambulatory status and gait patterns using multinomial logistic

Results: In our cohort 107 (71%) children were ambulant, of these 25 (16%) were aided and 43 (29%) non-ambulant. Of children who were ambulant 57 (53%) had bilateral spasticity, 37(35%) unilateral spasticity, 4 (4%) dystonia, 8 (7%) ataxia and 1 (1%) hypotonia. Gait Patterns were classified for bilateral spasticity as (true equinus=30, apparent equinus=2; jump knee=19; crouch=6) and for unilateral CP as (WGH I=4, II=29, III=1, IV=3). Quantitative global MRI severity scores (median 11, IQR=6.375–16.5) had a significant moderate correlation with children's ambulatory status (R^2 =0.138, p<0.001). Quantitative global MRI scores had a positive but non-significant association with severities of gait patterns in children with bilateral spasticity (R^2 =0.05, p=0.106) but no association with

regression (SPSS version 21).

gait patterns in unilateral CP. Qualitative classification (KM) of lesions did not correlate with ambulatory status (*p*=0.874).

Conclusions: Global severity scores of the brain lesion severity using a semi-quantitative MRI scale are related to ambulatory status in young children with CP. There was a trend towards an association between more severe gait patterns (crouch gait) as brain lesion severity increased in pre-school age children with CP. The ability to predict ambulatory status and gait pattern has potential to aid the streamlining of surveillance and tailoring of interventions based on early brain injury.

L6

Brain lesion severity and relationship to executive function in children with unilateral cerebral palsy

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Background/Objectives: To investigate the relationship between severity and location of brain lesions on structural MRI and executive functioning (EF) in children with unilateral cerebral palsy (UCP).

Design: Cross-sectional cohort study.

Participants and Setting: Of 52 children with hemiplegia (24 male; mean age 11.93 (SD 2.32) years; range 8 to 17 years, including GMFCS I=24, II=28; MACS 1=10, II=42; mean FSIQ(SD)=85.27 (17.3). Children were grouped according to brain lesion location: Predominantly right hemisphere (*n*=15), left hemisphere (*n*=23) and bilateral lesions (*n*=14).

Methods: Brain lesion severity was assessed using a reliable semi-quantitative scale (score 0–40; where higher score equates to greater lesion size). Lobar score (LS: frontal, parietal, temporal, occipital on each side), Hemispheric score (HS) ipsilateral to the lesion were calculated. Neuropsychological assessments (FSIQ) and four domains of executive function (EF) were assessed: attentional control; cognitive flexibility; goal setting; and information processing.

Results: The relationship between lesions size and IQ, EF were analysed using Pearson's correlations (SPSS 21). In unilateral lesions, left HS correlated with lower IQ r(23)=-0.692, p=0.000; poorer EF: on cognitive flexibility r(23)=-0.611, p=0.002; attentional control r(23)=-0.427, p=0.042; information processing r(23)=-0.646, p=0.001 and goal setting r(23)=-0.446, p=0.033. Right HS correlated with poorer information processing r(15)=-0.616, p=0.014 and with lower IQ scores r(15)=-0.589, p=0.021. In bilateral lesions: right HS correlated with lower IQ r(14)=-0.594, p=0.025 but not with EF domains. In unilateral lesions: left frontal LS correlated with poorer cognitive flexibility r(23)=-0.710, p=0.000, information processing r(23)=-0.612, p=0.002 and goal setting r(23)=-0.594, p=0.003. No correlation was found between right frontal LS and EF domains. For children with bilateral lesions: left frontal LS correlated with poorer information processing r (14)=-0.591, p=0.026; while the right frontal LS correlated with poorer goal setting tasks r(14)=-0.552, p=0.041.

Conclusions: Overall, for laterality of brain lesion (left, right, bilateral) there is a relationship between lower IQ scores and lesion severity. For children with left sided lesions there is a relationship between lesion size and executive dysfunction for the left frontal lobe and left hemisphere, not observed for children with right sided lesions (i.e., left hemiparesis). For bilateral lesions a relationship between executive dysfunction and greater lesion size was observed in the frontal regions, but not either hemisphere. This has implications for the planning of interventions and monitoring of child development as lesion location and size can potentially help to predict and inform interventions to improve Executive Function in children with UCP.

L7

Validation of a semi-quantitative scale for brain structural MRI in unilateral cerebral palsy: relationship with paretic hand sensorimotor function

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Background/Objectives: To examine the relationship between MRI quantitative brain structure and upper limb sensorimotor function in children with Unilateral Cerebral Palsy (UCP). Design: To examine the relationship between MRI quantitative brain structure and upper limb sensorimotor function in children with Unilateral Cerebral Palsy (UCP).

Participants and Setting: Of 71 children with UCP (35 left/36 right; GMFCS I=41, II=30; MACS I=32, II=39; aged 5–17 years) were recruited from a tertiary centre.

Methods: Children undertook 3T structural MRI which were scored on a novel reliable semi-quantitative scale (Fiori). Total Hemispheric (HS) and hemispheric summary scores (HSS with PLIC and basal ganglia (BG) ipsilateral to the lesion and the summary BG and brainstem (sBG & BS with PLIC and BG included) and global severity scores (GS) were calculated. Children were classified on House scale (1–9), assessed for unimanual capacity on Jebsen-Taylor Hand Function Test (JTHFT, seconds); Melbourne Assessment of Unilateral Upper Limb Function (MUUL) for impaired arm and bimanual coordination using Assisting Hand Assessment (AHA). Impaired hand sensation was assessed stereognosis (0–9) and 2-point-discrimination (2PD, mm). The relationship between HS, HSS, SBG & BSS and GS against sensorimotor measures were analysed using Pearson one-tailed, p<0.05 (SPSSv21).

Results: All the brain lesion severity scores showed an association with sensory-motor measures. HS had weak correlations (p<0.05) with House (r=0.27), JTHFT (r=0.22), MUUL (r=0.25) and 2PD (r=0.22). HSS had moderate correlations (p<0.01) with House (r=0.33), JTHFT(r=0.36), MUUL (r=0.37), AHA (r=0.35) and stereognosis (r=0.37), and weak correlation with 2PD (r=0.28; p=0.005). The sBG & BS had moderate correlations (p<0.01) with House (r=0.32), JTHFT (r=0.46), MUUL (r=0.47), AHA (r=50) and stereognosis

(r=0.43); and weak correlation (p<0.05) with 2PD (r=0.288). The GS had weak correlations (p<0.05) with House (r=0.23), JTHFT (r=0.21) and stereognosis (r=0.24), and moderate correlation with 2PD (r=0.326; p<0.01). The unilateral lesion severity score (HS) correlated strongly with motor outcome while the GS and sBG & BS correlated strongly with sensory measures. When BG measures are included in HSS there was a stronger correlation with sensory measures.

Conclusions: Our novel semi-quantitative scale for brain structural MRI exhibits good validity with regards to paretic hand sensorimotor function in a cohort of children with UCP. This quantitative structural MRI scoring is a useful clinical tool for studying brain structure-function relationships. Paretic hand motor function depends more on ipsilesional hemispheric brain lesion severity and vertical pattern involvement (lesion depth), while impaired sensory function depends on whole brain and basal ganglia (BG) lesion severity and transversal pattern involvement (lesion extent).

L8

Disruption of cerebro-cerebellar pathways in congenital unilateral brain lesions and correlation with hand function: a diffusion tractography study

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Background/Objectives: In adults with chronic unilateral stroke, a disruption of functional and structural connectivity between cerebrum and cerebellum (crossed cerebellar diaschis – CCD) has been correlated with paretic hand motor function. This relationship has not been studied in children with congenital unilateral brain lesions (UCP). This study examined the hypothesis that (1) structural cortico-ponto-cerebellar (CPC) connectivity is impacted by brain damage and (2) the degree of disruption is correlated with hand motor function.

Design: Case-control study.

Participants and Setting: Of 37 children with congenital UCP, no epilepsy (22 left sided lesion, age range 5–16 years, MACS I=13, II=24, GMFCS I=23, II=14 and compared to 20 typically developing children (age range 7–16 years) were enrolled from the community.

Methods: Magnetic resonance imaging structural connectivity at 3T was measured using high-angular-resolution diffusion imaging (HARDI) and region of interest (ROI) based probabilistic tractography. Connectivity of CPC tract was determined using asymmetry index (AI) based on the number of streamlines contained within the generated descending projections. Impaired hand function was tested using the Assisting Hand Assessment (AHA), the Melbourne assessment of Unilateral Upper Limb Function (MUUL) and Jebsen Taylor Test of Hand Function (JTTHF). Two-tailed t-test determined if the AI differed between cases and controls. Relationship between the AI and hand function measures was determined using

Pearson product-moment correlation. Analyses were repeated for the sub-groups of cortical and deep gray matter (CDGM, n=11) and periventricular white matter (PWM, n=18).

Results: The AI of CPC tracts was significantly different between UCP and controls (mean AI cases 0.58, SD 0.28; mean AI controls 0.23, SD 0.48; p=0.002), and for both subgroups CDGM (p=0.01) and PWM (p=0.01) independently, versus controls, but there were no differences between the subgroups. The AI were significantly related to AHA (r=33, p=0.04), with a coefficient of determination r² of 0.11. The AI in the PWM lesioned subgroup was moderately correlated with AHA (r=0.40; r²=0.16; p=0.03), but the correlation of AI and AHA scoring was not significant in the CDGM subgroup. No correlation was found between AI measures and unimanual capacity (MUUL or JTTHF) for any group.

Conclusions: This is the first study demonstrating marked reduction of structural connectivity in the cortico-ponto-cerebellar, CPC tract connecting the lesioned cerebral and the contralateral cerebellar hemispheres. The degree of connectivity disruption was correlated with AHA scores, supporting a key role of CPC in bimanual task coordination. Crossed cerebellar diaschis, CCD is present in children with UCP in the chronic phase and influences motor hand function. Further studies are needed to clarify the contribution of CCD on brain plasticity after a congenital brain lesion.

L9

Validation of a semi-quantitative structural MRI scale against hemispheric connections using a whole brain diffusion approach in children with unilateral cerebral palsy

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Background/Objectives: The study evaluates validity of a novel reliable semi-quantitative scale for brain structural magnetic resonance imaging (sMRI) using brain structural diffusion connectivity (dMRI) to assess disruption of connections in children with unilateral cerebral palsy (UCP).

Design: Cross sectional study.

Participants and Setting: Of 73 children with UCP (34 left/39 right; 40 males, GMFCS I=35/II=36 MACS I=32/II=39; Mean Age 10.5, SD 3.0years) were assessed.

Methods: High-resolution structural MRI at 3T were classified according to Krageloh-Mann qualitative system (KM) and scored on a semi-quantitative scoring system (score 0–40, Fiori). Probabilistic tractography was used to determine structural connections (dMRI) of between 73 cortical and subcortical regions. Fractional anisotropy (FA) and mean diffusivity (MD) were calculated as microstructure measures within each connection. The relationship between hemispheric summary score (HSS) and lobar score (LS) (Frontal, Parietal, Temporal, Occipital) for each hemisphere (right and left) on sMRI and differences in FA and MD of brain connections on

dMRI was assessed using a General Linear Model (GLM) (p-value defined as <0.05).

Results: Our sample included 50 children with PWM and 23 with CDGM lesions. HSS and LS for the hemisphere contralateral to the clinical presentation of hemiplegia showed no differences for right and left hemiplegia. As expected in right hemiplegia, the left HSS was correlated with a reduction in FA values (p=0.007) in a number of ipsilesional and cortico-cortical tracts and cortico-subcortical tracts. In left hemiplegia, the right HSS was correlated with a reduction in FA values (p<0.001), in a number of ipsilesional and interhemispheric cortico-cortical and cortico-subcortical tracts. In right hemiplegia, left LS was associated with a reduction in FA (p<0.01) in the frontal, temporal and occipital but not in the parietal lobe. In right hemiplegia, left LS was associated with a reduction in FA (p<0.001) in the frontal, temporal, parietal and occipital lobes.

Conclusions: Our novel semi-quantitative scale for structural MRI demonstrated a strong relationship with disruption of structural brain connectivity assessed by probabilistic tractography. The strongest relationships were between the HSS on the side of the brain lesion including the basal ganglia and PLIC with disruption to connectivity (decreased FA). Our data supports asymmetrical microstructural disruption between right and left hemispheres according to the side of hemiplegia, independently of the severity of lesion on structural MRI. Further studies on more homogeneous samples may determine the impact of dominance on secondary reorganisation post injury.

L10

Relationship between brain lesion severity and motor outcomes in pre-school aged children with cerebral palsy

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Background/Objectives: To examine the relationship between the severity of brain lesions on structural MRI (semi-quantitative scale) and prospectively collected motor outcomes in a pre-school aged cohort of children with CP.

Design: Prospective population-based cohort study.

Participants and Setting: Of 152 children with CP (57.9% male, 41.4% born preterm), classified as GMFCS I=70 (46.1%); II=14 (9.2%); III=25 (16.4%); IV=20 (13.2%); V=23 (15.1%); born between 1st January 2006 to 31st December 2009 were recruited from a population based sample.

Methods: Structural MRI neuroimaging (T1, T2, Flair) was performed at a mean age of 22.5 months (SD 17.0mo) and brain lesions were classified using two systems: (1) qualitative system developed by Krageloh-Mann in which lesions were placed into 1 of 5 mutually exclusive nominal categories according to the primary pattern of pathology, and (2) a novel quantitative scale which assigned each scan a score from 0 to 40 with higher scores indicating more extensive pathology (Fiori). All children underwent prospective motor assessment with GMFM-66, and were classified according to GMFCS, MACS and motor distribution at a mean age of 36.9 months (SD 5.1mo). The relationship between brain lesion severity scores and motor capacity was evaluated using linear regression analysis (GMFM-66) and multinomial logistic regression analysis (GMFCS and MACS).

Results: Quantitative MRI scores were significantly associated with a significant portion of variance in GMFM-66 scores (R^2 =0.26, p<0.01), GMFCS levels (pseudo R^2 =0.09, p<0.01), and MACS levels (pseudo R^2 =0.10 p<0.01). Specifically, as quantitative scale scores increased, GMFM-66 scores decreased, and GMFCS and MACS functional classification levels increased. The asymmetry indices of the hemispheric sub-scores on the quantitative scale were strongly associated with CP motor distribution (unilateral or bilateral). The quantitative MRI scale had a stronger association with GMFM-66 motor outcomes (R^2 =0.26) than the currently used qualitative MRI classification system (R^2 =0.05).

Conclusions: A novel quantitative MRI scale that is clinically accessible provides an objective description of the static heterogeneous brain lesions that define CP. This new MRI scale has strong potential in predicting the future gross motor capacity, manual ability classification and motor distribution of children with CP from around 2 years of age. As most children with CP receive MRI by 2 years of age our quantitative MRI scale has the potential to determine the relationship to later motor outcomes and manual abilities enabling the early tailoring of interventions and screening for prevention of complications.