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NEWS
Cerebral Palsy Research

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Interventions and Management

1. Efficacy of the small step program in a randomised controlled trial for infants below age 12 months with clinical signs of CP; a study protocol.

Eliasson AC, Holmström L, Aarne P, Nakeva von Mentzer C, Weiland AL, Sjöstrand L, Forssberg H, Tedroff K, Löwing K.

BMC Pediatr. 2016 Nov 3;16(1):175.

BACKGROUND: Children with cerebral palsy (CP) have life-long motor disorders, and they are typically subjected to extensive treatment throughout their childhood. Despite this, there is a lack of evidence supporting the effectiveness of early interventions aiming at improving motor function, activity, and participation in daily life. The study will evaluate the effectiveness of the newly developed Small Step Program, which is introduced to children at risk of developing CP during their first year of life. The intervention is based upon theories of early learning-induced brain plasticity and comprises important components of evidence-based intervention approaches used with older children with CP. METHOD AND DESIGN: A two-group randomised control trial will be conducted. Invited infants at risk of developing CP due to a neonatal event affecting the brain will be randomised to either the Small Step Program or to usual care. They will be recruited from Astrid Lindgren Children's Hospital at regular check-up and included at age 3-8 months. The Small Step Program was designed to provide individualized, goal directed, and intensive intervention focusing on hand use, mobility, and communication in the child's own home environment and carried out by their parents who have been trained and coached by therapists. The primary endpoint will be approximately 35 weeks after the start of the intervention, and the secondary endpoint will be at 2 years of age. The primary outcome measure will be the Peabody Developmental Motor Scale (second edition). Secondary assessments will measure and describe the children's general and specific development and brain pathology. In addition, the parents' perspective of the program will be evaluated. General linear models will be used to compare outcomes between groups. DISCUSSION: This paper presents the background and rationale for developing the Small-Step Program and the design and protocol of a randomized controlled trial. The aim of the Small Step Program is to influence development by enabling children to function on a higher level than if not treated by the program and to evaluate whether the program will affect parent's ability to cope with stress and anxiety related to having a child at risk of developing CP.

PMID: 27809886

2. Development, and construct validity and internal consistency of the Grasp and Reach Assessment of Brisbane (GRAB) for infants with asymmetric brain injury.

Perez M, Ziviani J, Guzzetta A, Ware RS, Tealdi G, Burzi V, Boyd RN.

Infant Behav Dev. 2016 Oct 27;45(Pt A):110-123. doi: 10.1016/j.infbeh.2016.10.004. [Epub ahead of print]

INTRODUCTION: Infants with asymmetric brain injury (asymBI) are at high risk of Unilateral Cerebral Palsy (UCP). The Grasp and Reach Assessment of Brisbane (GRAB) was developed to detect asymmetries in unimanual/bimanual upper limb

(UL) reach and grasp behaviours in infants with asymBI. This study reports the development of the GRAB and evaluates its construct validity and internal consistency. MATERIAL AND METHODS: Prospective study of twenty four infants with asymBI and twenty typically developing (TD) infants at 18 weeks corrected age (C.A.) in a structured play session. Three different coloured toys were presented at the midline in a block design of six 30-s trials of toy presentation, separated by five 30-s trials of no toy presentation. The number and duration of: (i) unimanual contacts; (ii) unimanual grasps; (iii) bimanual midline grasps; and (iv) duration of other unimanual behaviours (e.g. prehensile movements and transport phase) were measured. An Asymmetry Index (AI) was calculated to determine asymmetries between ULs. Possible AI values ranged from 0 to 100%, indicating proportion of toy presentation time that unimanual behaviours were asymmetric between ULs. Internal consistency of both the Time Phase (TP) and Toy Colour Phase (TCP) test items were determined by calculating Cronbach's alpha coefficients. Each assessment occasion was split into six TPs and two TCPs; whereby one TP comprised one 30-s trial of one toy presentation and one TCP comprised two 30-s trials of the same toy presentation. RESULTS: For TP, seven out of nine unimanual behaviours and two out of three bimanual behaviours demonstrated strong internal consistency (Cronbach's alpha coefficients 0.72-0.89). No unimanual activity demonstrated the strongest IC (0.89). For TCP, six out of nine unimanual behaviours demonstrated strong IC (0.73-0.82). Number of unimanual contacts and duration of unimanual prehensile movements demonstrated the strongest IC (0.82). Duration of unimanual contribution to hands at midline and duration of bimanual midline behaviour demonstrated the weakest IC for both TP and TCP (0.46-0.50). For unimanual contacts, the asymBI group were more asymmetric between ULs (mean AI=50%) compared to the TD group (mean AI=30%). For unimanual grasps, both groups were similarly asymmetric (both mean AI=40%). The TD group were almost twice as likely to demonstrate bimanual grasps as the asymBI group (incidence rate ratio IRR 1.9, 95% CI 1.4 to 2.5, $p<0.001$). Infants with asymBI were less likely to use the impaired UL compared to the unimpaired UL for grasping (IRR 0.6, 95% CI 0.5 to 0.8, $p<0.001$); and used the impaired UL for a shorter proportion of time compared to the unimpaired UL for grasping (mean difference -9.1%, 95% CI -16.6 to -1.7, $p=0.02$). CONCLUSIONS: The GRAB is a criterion-referenced research measure that detects and quantifies the presence or absence of unimanual and bimanual reach and grasp behaviours at 18 weeks C.A. in infants at risk of UCP. The GRAB demonstrated moderate to strong construct validity and strong IC within an assessment occasion. There was no toy preference or warm-up effect for TP or TCP for either group; confirming that the GRAB is a consistent measure across toy presentations within an assessment occasion. In this study, the GRAB identified that infants with asymBI demonstrated a paucity of bimanual grasping compared to TD infants; and demonstrated asymmetric unimanual grasping between ULs at 18 weeks C.A.

PMID: 27810684

3. Management of the spastic hip in cerebral palsy.

Givon U.

Curr Opin Pediatr. 2016 Oct 27. [Epub ahead of print]

PURPOSE OF REVIEW: Spastic hip dysplasia (SHD) is a common finding in patients with cerebral palsy, with a higher incidence in more involved patients, causing disability and reducing quality of life in these patients. SHD is the most serious orthopedic problem seen in cerebral palsy patients, and requires special attention and tenacious evaluation of the patients. The aim of this article is to review the new developments in the treatment of SHD. RECENT FINDINGS: Patients with cerebral palsy were shown to have better hip joint morphology when they had access to hip surveillance programmes, with proactive search of patients with progressing hip subluxation and early intervention. Prediction of progression of SHD is now available based on the experience of these programmes.Patients who underwent hip joint reconstruction showed that incongruent joints remodeled following a Dega osteotomy. Patients who underwent a varus osteotomy of the femoral neck without pelvic reconstruction had a higher rate of recurrence when they were older and the SHD was more severe. Health-related quality of life measures improved following hip joint reconstructions and salvage procedures. CONCLUSION: Patients with cerebral palsy should be monitored with a well-defined hip surveillance programme, with early identification and timely intervention for SHD.

PMID: 27798427

4. Long-term changes in femoral anteversion and hip rotation following femoral derotational osteotomy in children with cerebral palsy.

Boyer E, Novacheck TF, Rozumalski A, Schwartz MH.

Gait Posture. 2016 Oct;50:223-228. doi: 10.1016/j.gaitpost.2016.09.004. Epub 2016 Sep 8.

BACKGROUND: Excessive femoral anteversion is common in cerebral palsy (CP), is often associated with internal hip rotation during gait, and is frequently treated with a femoral derotational osteotomy (FDO). Concerns exist regarding long-term maintenance of surgical outcomes. Past studies report varying rates of recurrence, but none have employed a control group. METHODS: We conducted a retrospective analysis examining long-term (~5 years) changes in anteversion and hip rotation following FDO in children with CP. We included a control group that was matched for age and exhibited excessive anteversion (>30°) but did not undergo an FDO. Anteversion, mean stance hip rotation, and rates of problematic remodeling and recurrence were assessed (>15° change and final level outside of normal limits). RESULTS: The control group was reasonably well matched, but exhibited 9° less anteversion and 3° less internal hip rotation at the pre time point. At a five year follow-up, the FDO group had less anteversion than the control group (20° vs. 35°, $p<0.05$). The mean stance phase hip rotation did not differ between the groups (4° vs. 5°, $p=0.17$). Over one third of limbs remained excessively internal in both groups (FDO: 34%, Control: 37%). Rates of problematic recurrence and remodeling were low (0%-11%). CONCLUSIONS: An FDO is an effective way to correct anteversion in children with CP. Long-term hip rotation is not fully corrected by the procedure, and is not superior to a reasonably well matched control group. Rates of problematic recurrence and remodeling are low, and do not differ between the groups.

PMID: 27653149

5. A cohort study of tibialis anterior tendon shortening in combination with calf muscle lengthening in spastic equinus in cerebral palsy.

Tsang ST, McMorran D, Robinson L, Herman J, Robb JE, Gaston MS.

Gait Posture. 2016 Oct;50:23-27. doi: 10.1016/j.gaitpost.2016.08.015. Epub 2016 Aug 17.

The aim of this study was to evaluate the outcome of combined tibialis anterior tendon shortening (TATS) and calf muscle-tendon lengthening (CMTL) in spastic equinus. Prospectively collected data was analysed in 26 patients with hemiplegic (n=13) and diplegic (n=13) cerebral palsy (CP) (GMFCS level I or II, 14 males, 12 females, age range 10-35 years; mean 16.8 years). All patients had pre-operative 3D gait analysis and a further analysis at a mean of 17.1 months (±5.6months) after surgery. None was lost to follow-up. Twenty-eight combined TATS and CMTL were undertaken and 19 patients had additional synchronous multilevel surgery. At follow-up 79% of patients had improved foot positioning at initial contact, whilst 68% reported improved fitting or reduced requirement of orthotic support. Statistically significant improvements were seen in the Movement Analysis Profile for ankle dorsi-/plantarflexion (4.15°, $p=0.032$), maximum ankle dorsiflexion during swing phase (11.68°, $p<0.001$), and Edinburgh Visual Gait Score (EVGS) (4.85, $p=0.014$). Diplegic patients had a greater improvement in the EVGS than hemiplegics (6.27 -vs- 2.21, $p=0.024$). The originators of combined TATS and CMTL showed that it improved foot positioning during gait. The present study has independently confirmed favourable outcomes in a similar patient population and added additional outcome measures, the EVGS, foot positioning at initial contact, and maximum ankle dorsiflexion during swing phase. Study limitations include short term follow-up in a heterogeneous population and that 19 patients had additional surgery. TATS combined with CMTL is a recommended option for spastic equinus in ambulatory patients with CP.

PMID: 27559938

6. Astym Therapy Improves Bilateral Hamstring Flexibility and Achilles Tendinopathy in a Child with Cerebral Palsy: A Retrospective Case Report.

Scheer NA, Alstat LR, Van Zant RS.

Clin Med Insights Case Rep. 2016 Oct 20;9:95-98. eCollection 2016.

PURPOSE: The purpose of this case report was to describe the use of Astym therapy to improve hamstring flexibility and Achilles tendinopathy in a child with cerebral palsy. CASE DESCRIPTION: An eight-year-old female with cerebral palsy was referred to physical therapy for the treatment of bilateral hamstring inflexibility and Achilles tendinopathy. Treatment focused

on an Astym therapy protocol of eccentric exercise, stretching, active and passive range of motion, gait training, and a home exercise program. The patient underwent a total of 11 physical therapy treatment sessions. OUTCOMES: At the conclusion of treatment, the patient demonstrated improved resting muscle tone in bilateral lower extremities with active 90/90 hamstring flexibility measured at 165° and ankle dorsiflexion active range of motion of 5° without pain at 0° and 90° knee flexion. The patient exhibited an improved gait pattern with even stride length and diminished genu recurvatum, decreased pain with standing and walking, discontinued use of ankle-foot orthoses, and improved activity tolerance and overall function for daily activities. DISCUSSION: The results of this case report indicate that physical therapy rehabilitation utilizing an Astym therapy protocol can successfully achieve gains in flexibility and strength and allow for improved function of bilateral lower extremities in a patient with cerebral palsy. CONCLUSION: Based on the findings of this case report, clinicians should consider the use of Astym therapy in treating musculoskeletal soft tissue dysfunction in pediatric patients with cerebral palsy.

PMID: 27790051

7. Feasibility and reliability of using an exoskeleton to emulate muscle contractures during walking.

Attias M, Bonnefoy-Mazure A, De Coulon G, Cheze L, Armand S.

Gait Posture. 2016 Oct;50:239-245. doi: 10.1016/j.gaitpost.2016.09.016. Epub 2016 Sep 19.

Contracture is a permanent shortening of the muscle-tendon-ligament complex that limits joint mobility. Contracture is involved in many diseases (cerebral palsy, stroke, etc.) and can impair walking and other activities of daily living. The purpose of this study was to quantify the reliability of an exoskeleton designed to emulate lower limb muscle contractures unilaterally and bilaterally during walking. An exoskeleton was built according to the following design criteria: adjustable to different morphologies; respect of the principal lines of muscular actions; placement of reflective markers on anatomical landmarks; and the ability to replicate the contractures of eight muscles of the lower limb unilaterally and bilaterally (psoas, rectus femoris, hamstring, hip adductors, gastrocnemius, soleus, tibialis posterior, and peroneus). Sixteen combinations of contractures were emulated on the unilateral and bilateral muscles of nine healthy participants. Two sessions of gait analysis were performed at weekly intervals to assess the reliability of the emulated contractures. Discrete variables were extracted from the kinematics to analyse the reliability. The exoskeleton did not affect normal walking when contractures were not emulated. Kinematic reliability varied from poor to excellent depending on the targeted muscle. Reliability was good for the bilateral and unilateral gastrocnemius, soleus, and tibialis posterior as well as the bilateral hamstring and unilateral hip adductors. The exoskeleton can be used to replicate contracture on healthy participants. The exoskeleton will allow us to differentiate primary and compensatory effects of muscle contractures on gait kinematics.

PMID: 27665088

8. [The regulation of balance in the children presenting with severe cerebral palsy following the treatment with the use of the locomotor training in combination with the electrical stimulation of leg muscles and spinal cord].

[Article in Russian]

Nikityuk IE, Moshonkina TR, Gerasimenko YP, Vissarionov SV, Baindurashvili AG.

Vopr Kurortol Fizioter Lech Fiz Kult. 2016;93(5):23-27.

One of the cerebral palsy (CP) symptoms is the impairment of the ability to maintain upright standing. The objective of the present study was to investigate the effect of the locomotor training combined with electrical transcutaneous spinal cord stimulation (TSCS) and functional electrical stimulation (FES) of the muscles to facilitate the restoration of the vertical posture function in the children presenting with cerebral palsy. PATIENTS AND METHODS: This article is designed to present the results of the study that included 19 patients at the age from 6 to 12 years. The severity of the CP clinical manifestations was estimated to be around 3 in accordance with The Gross Motor Function Classification System (GMFCS). All the patients underwent 15 half-hour daily sessions of robotic mechanotherapy with the use of a locomotor training device. In 8 patients from the main group, each locomotor training session was accompanied by the TSCS and FES procedures. The patients of the control group underwent the locomotor training alone. RESULTS: The dynamics of regaining the standing posture regulation was evaluated by stabilometry that has revealed a significant increase of postural stability in the children of the main group in comparison with those in the control group. It was apparent as a statistically significant ($p<0.05$) reduction of the length and the area of the center of pressure (COP) projection with open eyes (the 167 mm and 112 mm2 decrease of the median, respectively). In addition, the tendency toward the normalization of the COP projection in the sagittal plane was documented.

CONCLUSION: The present study has demonstrated the normalization of the impaired balance control system in the children presenting with the severe form of cerebral palsy under the influence of the combined treatment with the use of transcutaneous spinal cord stimulation and functional electrical stimulation of legs muscles supplemented by the locomotor training.

PMID: 27801408

9. Subclassification of GMFCS Level-5 Cerebral Palsy as a Predictor of Complications and Health-Related Quality of Life After Spinal Arthrodesis.

Jain A, Sponseller PD, Shah SA, Samdani A, Cahill PJ, Yaszay B, Njoku DB, Abel MF, Newton PO, Marks MC, Narayanan UG; Harms Study Group.

J Bone Joint Surg Am. 2016 Nov 2;98(21):1821-1828.

BACKGROUND: The Gross Motor Function Classification System (GMFCS) of cerebral palsy categorizes patients by mobility. Patients at GMFCS level 5 are considered the most disabled and at high risk of hip and spine problems, yet they represent a wide spectrum of function. Our aim was to subclassify patients at GMFCS level 5 who underwent spinal arthrodesis on the basis of central neuromotor impairments and to assess whether subclassification predicted postoperative complications and changes in health-related quality of life. METHODS: Using a prospective cerebral palsy registry, we identified 199 patients at GMFCS level 5 who underwent spinal arthrodesis from 2008 to 2013. Patients were assigned to subgroups according to preoperative central neuromotor impairments: the presence of a gastrostomy tube, a tracheostomy, history of seizures, and nonverbal status. Nine percent of patients had 0 impairments (GMFCS level 5.0), 14% had 1 impairment (level 5.1), 26% had 2 impairments (level 5.2), and 51% had 3 or 4 impairments (level 5.3). The Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD) questionnaire was used for preoperative and postoperative health-related quality-of-life outcome assessments, and major complications were recorded. RESULTS: The rate of major complications increased significantly with higher GMFCS level-5 subtype ($p = 0.002$), with 12% at level 5.0, 21% at level 5.1, 31% at level 5.2, and 49% at level 5.3. Five of the 7 patients who died within the follow-up period were at level 5.3. No significant differences were found among subgroups with respect to the magnitude of correction of the major coronal curve or pelvic obliquity. Preoperative and final follow-up CPCHILD total scores decreased significantly from GMFCS level 5.0 to level 5.3. However, no significant differences were found by subgroup with respect to the magnitude of improvement in CPCHILD total scores from the preoperative to the final follow-up evaluation ($p = 0.597$). CONCLUSIONS: Stratification based on central neuromotor impairments can help to identify patients with cerebral palsy at GMFCS level 5 who are at higher risk for developing complications after spinal arthrodesis.

PMID: 27807115

10. Hypopituitarism in children with cerebral palsy.

Uday S, Shaw N, Krone R, Kirk J.

Arch Dis Child. 2016 Oct 27. pii: archdischild-2016-311012. doi: 10.1136/archdischild-2016-311012. [Epub ahead of print]

Poor growth and delayed puberty in children with cerebral palsy is frequently felt to be related to malnutrition. Although growth hormone deficiency is commonly described in these children, multiple pituitary hormone deficiency (MPHD) has not been previously reported. We present a series of four children with cerebral palsy who were born before 29 weeks gestation who were referred to the regional endocrinology service, three for delayed puberty and one for short stature, in whom investigations identified MPHD. All patients had a height well below -2 standard deviation score (2nd centile) at presentation and three who had MRI scans had an ectopic posterior pituitary gland. We therefore recommend that the possibility of MPHD should be considered in all children with cerebral palsy and poor growth or delayed puberty. Early diagnosis and treatment is essential to maximise growth and prevent associated morbidity and mortality.

PMID: 27789461

11. Clinical usefulness of brain-computer interface-controlled functional electrical stimulation for improving brain activity in children with spastic cerebral palsy: a pilot randomized controlled trial.

Kim TW, Lee BH.

J Phys Ther Sci. 2016 Sep;28(9):2491-2494. Epub 2016 Sep 29.

[Purpose] Evaluating the effect of brain-computer interface (BCI)-based functional electrical stimulation (FES) training on brain activity in children with spastic cerebral palsy (CP) was the aim of this study. [Subjects and Methods] Subjects were randomized into a BCI-FES group (n=9) and a functional electrical stimulation (FES) control group (n=9). Subjects in the BCI-FES group received wrist and hand extension training with FES for 30 minutes per day, 5 times per week for 6 weeks under the BCI-based program. The FES group received wrist and hand extension training with FES for the same amount of time. Sensorimotor rhythms (SMR) and middle beta waves (M-beta) were measured in frontopolar regions 1 and 2 (Fp1, Fp2) to determine the effects of BCI-FES training. [Results] Significant improvements in the SMR and M-beta of Fp1 and Fp2 were seen in the BCI-FES group. In contrast, significant improvement was only seen in the SMR and M-beta of Fp2 in the control group. [Conclusion] The results of the present study suggest that BCI-controlled FES training may be helpful in improving brain activity in patients with cerebral palsy and may be applied as effectively as traditional FES training.

PMID: 27799677

12. Pain report and musculoskeletal impairment in young people with severe forms of cerebral palsy: A population-based series.

McDowell BC, Duffy C, Lundy C.

Res Dev Disabil. 2016 Oct 25. pii: S0891-4222(16)30226-8. doi: 10.1016/j.ridd.2016.10.006. [Epub ahead of print]

BACKGROUND: While pain is reportedly more prevalent in more functionally impaired children with cerebral palsy, information is scant in those with poor communication skills. METHODS: Young people (4-27 years) with severe forms of cerebral palsy were recruited from a population-based register. The Child Health Questionnaire (CHQ) provided information on general health and bodily pain; the Paediatric Pain Profile (PPP) was used for participants with limited communication; and the Spinal Alignment and Range of Motion Measure (SAROMM) described musculoskeletal impairment. RESULTS: 123 young people (GMFCS IV=55 and V=68) and their families/carers participated. Fourteen percent of CHQ responses (n=123) reported severe/very severe pain in recent weeks, whilst 7% reported pain every/almost every day. CHQ pain report was significantly higher for young people in GMFCS level V and correlated significantly with both global health and musculoskeletal impairment. High levels of pain were recorded on the PPP for non-communicating children but only a weak correlation between PPP and CHQ scores was detected. CONCLUSION: Managing pain in young people with severe musculoskeletal and cognitive impairment presents a huge challenge to carers and professionals. The PPP may represent a useful adjunct in those young people with severe communication difficulties.

PMID: 27793550

13. The effects of connective tissue manipulation and Kinesio Taping on chronic constipation in children with cerebral palsy: a randomized controlled trial.

Orhan C, Kaya Kara O, Kaya S, Akbayrak T, Kerem Gunel M, Baltaci G.

Disabil Rehabil. 2016 Oct 28:1-11. [Epub ahead of print]

PURPOSE: The aim of this study was to investigate the effects of connective tissue manipulation (CTM) and Kinesio Taping® (KT) on constipation and quality of life in children with cerebral palsy (CP). METHOD: This study was designed as a randomized controlled trial. Forty children diagnosed with chronic constipation based on Rome III criteria were randomly assigned to CTM group [6 females, 7 males; 8 y 6 mo (SD = 3y 4 mo)], KT group [7 female, 7 male; 8y 7 mo (SD =3y 5 mo)] or control group [6 female, 7 male; 8y 3 mo (SD = 3y 6 mo)]. All patients were assessed with 7-day bowel diaries, Bristol Stool Form Scale (BSFS), Visual Analog Scale (VAS), and Pediatric Quality of Life Inventory (PEDsQL). Kruskal-Wallis, Wilcoxon's signed-rank, and Mann-Whitney U tests were used to determine intra-group and inter-group differences. The level of significance was $p < 0.05$. RESULTS: Among the CTM, KT, and control groups, there were statistically significant

differences regarding the changes in defecation frequency (2.46, 3.00, 0.30, ES 1.16, p < 0.001), duration of defecation (5.07, 5.35, 0.15, ES 2.37, p = 0.003), BSFS (1.84, 2.14, 0.07, ES 0.91, p < 0.001), VAS (4.83, 3.87, 0.23, ES 1.98, p < 0.001), and PEDsQL total scores (7, 14, 8.36, -0.85, ES 4.08, p < 0.001). CONCLUSIONS: This study revealed that CTM and KT seem equally effective physiotherapy approaches for the treatment of pediatric constipation and these approaches may be added to bowel rehabilitation program. Implications for rehabilitation CTM and KT have similar effectiveness in alleviating the constipation-related symptoms and improving quality of life in children with CP. CTM and KT can be integrated into bowel rehabilitation programs. Considering the characteristics of patients, these treatment options can be used as an alternative of each other by physiotherapists.

PMID: 27793072

14. Parenting acceptance and commitment therapy: a randomised controlled trial of an innovative online course for families of children with cerebral palsy.

Whittingham K, Sheffield J, Boyd RN.

BMJ Open. 2016 Oct 19;6(10):e012807. doi: 10.1136/bmjopen-2016-012807.

INTRODUCTION: Cerebral palsy (CP) impacts on the entire family in a manner that is long-term, complex and multifactorial. In addition, the quality of the parent-child relationship impacts on many and varied child outcomes, making the provision of easily accessible and evidence-based support to parents of children with CP a priority. This paper reports the protocol of a randomised controlled trial of an innovative and translatable online intervention, parenting acceptance and commitment therapy (PACT), for families of children with CP. We predict that participating in the PACT programme will be associated with improvements in the parent-child relationship, in child functioning and in adjustment and quality of life for both parent and child. METHODS AND ANALYSIS: We aim to recruit 66 parents of children (2-10 years old) diagnosed with CP to this study. Families will be randomly assigned to two groups: wait-list control and PACT. PACT is a parenting intervention grounded in acceptance and commitment therapy (ACT) and developed into an online course 'PARENT101 Parenting with Purpose' using the edX platform. All participants will be offered PACT before completion of the study. Assessments will take place at baseline, following completion of PACT and at 6-month follow-up (retention) and will focus on the parent-child relationship, parent and child adjustment and parent and child quality of life. Analysis will follow standard methods for randomised controlled trials using general linear models, specifically analysis of variance or analysis of covariance. ETHICS AND DISSEMINATION: Ethics approvals have been obtained through the Children's Health Queensland Hospital and Health Service Human Research Ethics Committee (HREC/15/QRCH/115) and The University of Queensland (2015001743). If efficacy is demonstrated, then the PARENT101 course has the potential to be disseminated widely in an accessible manner and at minimal cost. Further, the PACT framework may provide a blueprint for similar online courses with parents in a full range of contexts.

PMID: 27798022

15. Health Related Quality of Life and Care Giver Burden Following Spinal Fusion in Children with Cerebral Palsy.

DiFazio R, Miller P, Vessey JA, Snyder B.

Spine (Phila Pa 1976). 2016 Oct 25. [Epub ahead of print]

STUDY DESIGN: Prospective longitudinal cohort OBJECTIVE.: Evaluate changes in caregivers' perceptions of health related quality of life (HRQOL) and caregiver burden in children with severe cerebral palsy (CP) following spinal fusion. SUMMARY OF BACKGROUND DATA: Progressive scoliosis is common in non-ambulatory children with CP; the utility of spine fusion has been long debated and prospective evaluations of patient reported outcomes are limited. METHODS: Children 3-21 years old, GMFCS IV-V CP, scheduled for spine fusion were enrolled consecutively from September 2011-March 2014. Caregivers completed the CPCHILD and ACEND pre-operatively and at 6 weeks, 3, 6, 12, and 24 months post-operatively. Changes in CPCHILD™ and ACEND scores from pre-operative to 1 and 2 years after surgery were assessed using paired t-tests. Correlations between pre-operative Cobb angle and CPCHILD™ and ACEND scores were evaluated using Pearson's correlation analysis. RESULTS: Twenty-six GMFCS IV-V CP patients with severe scoliosis treated with spine fusion were included. Mean age was 14 years, 50% male, 46% had instrumentation to the pelvis. Average pre-operative Cobb angle was 68.9° (SD 25.68) with an average improvement of 76%. The CPCHILD™ score increased by 9.8 points above baseline (95% CI: 3.4 to 16.2) at 1 year post-operatively (p=0.005). However, at 2 years, the CPCHILD™ score regressed to baseline (p= 0.40). ACEND scores did not change from baseline scores at 1 year (p=0.09) and 2 year (0.72) follow-up; reflecting that

caregiver burden is little changed by spine fusion. While there was no correlation between pre-operative Cobb angle and CPCHILD™ score (p=0.52) or ACEND score (p=0.56) at 1 year or 2 year follow-up (p=0.69, p=0.90). Children with Cob ≤ 75° experienced more improvement 1 year after surgery than children with Cobb >75°. CONCLUSIONS: HRQOL improves 1 year following spine fusion but regresses to baseline after 2 years. Caregiver burden was unchanged following spine fusion.

PMID: 27792122

16. Longitudinal development of communication in children with cerebral palsy between 24 and 53 months: Predicting speech outcomes.

Hustad KC, Allison KM, Sakash A, McFadd E, Broman AT, Rathouz PJ.

Dev Neurorehabil. 2016 Oct 28:1-8. [Epub ahead of print]

OBJECTIVE: To determine whether communication at 2 years predicted communication at 4 years in children with cerebral palsy (CP); and whether the age a child first produces words imitatively predicts change in speech production. METHOD: 30 children (15 males) with CP participated and were seen 5 times at 6-month intervals between 24 and 53 months (mean age at time 1 = 26.9 months (SD 1.9)). Variables were communication classification at 24 and 53 months, age that children were first able to produce words imitatively, single-word intelligibility, and longest utterance produced. RESULTS: Communication at 24 months was highly predictive of abilities at 53 months. Speaking earlier led to faster gains in intelligibility and length of utterance and better outcomes at 53 months than speaking later. CONCLUSION: Inability to speak at 24 months indicates greater speech and language difficulty at 53 months and a strong need for early communication intervention.

PMID: 27792399

17. Integrating semantic dimension into openEHR archetypes for the management of cerebral palsy electronic medical records.

Ellouze AS, Bouaziz R, Ghorbel H.

J Biomed Inform. 2016 Oct;63:307-324. doi: 10.1016/j.jbi.2016.08.018. Epub 2016 Aug 24.

PURPOSE: Integrating semantic dimension into clinical archetypes is necessary once modeling medical records. First, it enables semantic interoperability and, it offers applying semantic activities on clinical data and provides a higher design quality of Electronic Medical Record (EMR) systems. However, to obtain these advantages, designers need to use archetypes that cover semantic features of clinical concepts involved in their specific applications. In fact, most of archetypes filed within open repositories are expressed in the Archetype Definition Language (ALD) which allows defining only the syntactic structure of clinical concepts weakening semantic activities on the EMR content in the semantic web environment. This paper focuses on the modeling of an EMR prototype for infants affected by Cerebral Palsy (CP), using the dual model approach and integrating semantic web technologies. Such a modeling provides a better delivery of quality of care and ensures semantic interoperability between all involved therapies' information systems. METHODS: First, data to be documented are identified and collected from the involved therapies. Subsequently, data are analyzed and arranged into archetypes expressed in accordance of ADL. During this step, open archetype repositories are explored, in order to find the suitable archetypes. Then, ADL archetypes are transformed into archetypes expressed in OWL-DL (Ontology Web Language - Description Language). Finally, we construct an ontological source related to these archetypes enabling hence their annotation to facilitate data extraction and providing possibility to exercise semantic activities on such archetypes. RESULTS: Semantic dimension integration into EMR modeled in accordance to the archetype approach. The feasibility of our solution is shown through the development of a prototype, baptized "CP-SMS", which ensures semantic exploitation of CP EMR. This prototype provides the following features: (i) creation of CP EMR instances and their checking by using a knowledge base which we have constructed by interviews with domain experts, (ii) translation of initially CP ADL archetypes into CP OWL-DL archetypes, (iii) creation of an ontological source which we can use to annotate obtained archetypes and (vi) enrichment and supply of the ontological source and integration of semantic relations by providing hence fueling the ontology with new concepts, ensuring consistency and eliminating ambiguity between concepts. CONCLUSIONS: The degree of semantic interoperability that could be reached between EMR systems depends strongly on the quality of the used archetypes. Thus, the integration of semantic dimension in archetypes modeling process is crucial. By creating an ontological source and annotating archetypes, we create a supportive platform ensuring semantic interoperability between archetypes-based EMR-systems.

PMID: 27568295

18. Application of the International Classification of Functioning, Disability and Health - Children and Youth in Children With Cerebral Palsy.

Jeevanantham D.

Indian Pediatr. 2016 Sep 8;53(9):805-810. Epub 2016 Jun 1.

The International Classification of Functioning, Disability and Health (ICF) is a framework for describing health status; however, there is a gap in literature for supporting its use as a classification tool. The purpose of this paper is to provide a perspective on its use in describing children with cerebral palsy. The interconnected concepts of the ICF are more important than the classification elements itself. Further research is required to prove its use as a classification tool in clinical practice.

PMID: 27395840

Prevention and Cure

19. Funding for cerebral palsy research in Australia, 2000-2015: an observational study.

Herbert DL, Barnett AG, White R, Novak I, Badawi N.

BMJ Open. 2016 Oct 24;6(10):e012924. doi: 10.1136/bmjopen-2016-012924.

OBJECTIVES: To examine the funding for cerebral palsy (CP) research in Australia, as compared with the National Institutes of Health (NIH). DESIGN: Observational study. SETTING: For Australia, philanthropic funding from Cerebral Palsy Alliance Research Foundation (CPARF) (2005-2015) was compared with National Health and Medical Research Council (NHMRC, 2000-2015) and Australian Research Council (ARC, 2004-2015) and CPARF and NHMRC funding were compared with NIH funding (USA). PARTICIPANTS: Cerebral Palsy researchers funded by CPARF, NHMRC or NIH. RESULTS: Over 10 years, total CPARF philanthropic funding was $21.9 million, including people, infrastructure, strategic and project support. As competitive grants, CPARF funded $11.1 million, NHMRC funded $53.5 million and Australian Research Council funded $1.5 million. CPARF, NHMRC and NIH funding has increased in real terms, but only the NIH statistically significantly increased in real terms (mean annual increase US$4.9 million per year, 95% CI 3.6 to 6.2, $p<0.001$). The NHMRC budget allocated to CP research remained steady over time at 0.5%. A network analysis indicated the relatively small number of CP researchers in Australia is mostly connected through CPARF or NHMRC funding. CONCLUSIONS: Funding for CP research from the Australian government schemes has stabilised and CP researchers rely on philanthropic funding to fill this gap. In comparison, the NIH is funding a larger number of CP researchers and their funding pattern is consistently increasing.

PMID: 27798026

20. Preterm Hypoxic-Ischemic Encephalopathy.

Gopagondanahalli KR, Li J, Fahey MC, Hunt RW, Jenkin G, Miller SL, Malhotra A.

Front Pediatr. 2016 Oct 20;4:114. eCollection 2016.

Hypoxic-ischemic encephalopathy (HIE) is a recognizable and defined clinical syndrome in term infants that results from a severe or prolonged hypoxic-ischemic episode before or during birth. However, in the preterm infant, defining hypoxic-ischemic injury (HII), its clinical course, monitoring, and outcomes remains complex. Few studies examine preterm HIE, and these are heterogeneous, with variable inclusion criteria and outcomes reported. We examine the available evidence that implies that the incidence of hypoxic-ischemic insult in preterm infants is probably higher than recognized and follows a more complex clinical course, with higher rates of adverse neurological outcomes, compared to term infants. This review aims to elucidate the causes and consequences of preterm hypoxia-ischemia, the subsequent clinical encephalopathy syndrome, diagnostic tools, and outcomes. Finally, we suggest a uniform definition for preterm HIE that may help in identifying infants most at risk of adverse outcomes and amenable to neuroprotective therapies.

PMID: 27812521

21. Abnormal neurodevelopmental outcomes are very likely in cases of bilateral neonatal arterial ischaemic stroke.

Jin JH, Shin JE, Lee S, Eun HS, Park MS, Park KI, Namgung R.

Acta Paediatr. 2016 Nov 3. doi: 10.1111/apa.13655. [Epub ahead of print]

AIM: Neonatal arterial ischaemic stroke (AIS) is an important cause of severe neurological disability. This study aimed to analyse the clinical manifestations and outcomes of AIS patients. METHODS: We enrolled neonates with AIS admitted to Severance Children's Hospital and Gangnam Severance Hospital between 2008 and 2015. AIS was confirmed using magnetic resonance imaging (MRI). We retrospectively reviewed the clinical manifestations, MRI findings, electroencephalography (EEG) findings and neurodevelopmental outcomes. RESULTS: The study comprised 29 neonates (18 boys). The mean follow-up period was 15.4 months (range 6-44 months) and the mean age at diagnosis was 8.1 days. Seizure was the most common symptom (66%). Bilateral involvement was more common than unilateral involvement (52%). The middle cerebral artery was the most commonly identified territory (79%). Abnormal EEG findings were noted in 93% of the cases. Neurodevelopment was normal in 11 (38%) patients, while cerebral palsy and delayed development were noted in eight (28%) and six (21%) patients, respectively. Patients with bilateral involvement were very likely to have abnormal neurodevelopmental outcomes. CONCLUSION: Our study showed that abnormal neurodevelopmental outcomes were very likely after cases of neonatal AIS with bilateral involvement and clinicians should consider early and more effective interventions in such

PMID: 27809371

22. Identifying risk factors for cerebral palsy.

[No authors listed]

Randomised controlled trials of rigorous management of adverse antenatal factors on the frequency of cerebral palsy are needed, Oxford researchers say.

Nurs Stand. 1996 Jan 9;10(15):14.

PMID: 27809110

23. Maternal infection and cerebral palsy.

[No authors listed]

Intrauterine exposure to maternal infection is associated with a marked increase in the risk of cerebral palsy in infants with normal birth weight.

Nurs Stand. 1997 Aug 13;11(47):29.

PMID: 27801113

24. Effect of storage time on gene expression data acquired from unfrozen archived newborn blood spots.

Ho NT, Busik JV, Resau JH, Paneth N, Khoo SK.

Mol Genet Metab. 2016 Nov;119(3):207-213. doi: 10.1016/j.ymgme.2016.08.001. Epub 2016 Aug 18.

Unfrozen archived newborn blood spots (NBS) have been shown to retain sufficient messenger RNA (mRNA) for gene expression profiling. However, the effect of storage time at ambient temperature for NBS samples in relation to the quality of gene expression data is relatively unknown. Here, we evaluated mRNA expression from quantitative real-time PCR (qRT-PCR) and microarray data obtained from NBS samples stored at ambient temperature to determine the effect of storage time on the quality of gene expression. These data were generated in a previous case-control study examining NBS in 53 children with

cerebral palsy (CP) and 53 matched controls. NBS sample storage period ranged from 3 to 16years at ambient temperature. We found persistently low RNA integrity numbers (RIN=2.3±0.71) and 28S/18S rRNA ratios (~0) across NBS samples for all storage periods. In both qRT-PCR and microarray data, the expression of three common housekeeping genes-beta cytoskeletal actin (ACTB), glyceraldehyde 3-phosphate dehydrogenase (GAPDH), and peptidylprolyl isomerase A (PPIA)-decreased with increased storage time. Median values of each microarray probe intensity at log2 scale also decreased over time. After eight years of storage, probe intensity values were largely reduced to background intensity levels. Of 21,500 genes tested, 89% significantly decreased in signal intensity, with 13,551, 10,730, and 9925 genes detected within 5years, > 5 to <10years, and >10years of storage, respectively. We also examined the expression of two gender-specific genes (X inactivation-specific transcript, XIST and lysine-specific demethylase 5D, KDM5D) and seven gene sets representing the inflammatory, hypoxic, coagulative, and thyroidal pathways hypothesized to be related to CP risk to determine the effect of storage time on the detection of these biologically relevant genes. We found the gender-specific genes and CP-related gene sets detectable in all storage periods, but exhibited differential expression (between male vs. female or CP vs. control) only within the first six years of storage. We concluded that gene expression data quality deteriorates in unfrozen archived NBS over time and that differential gene expression profiling and analysis is recommended for those NBS samples collected and stored within six years at ambient temperature.

PMID: 27553879

25. White Matter Injury and General Movements in High-Risk Preterm Infants.

Peyton C, Yang E, Msall ME, Adde L, Støen R, Fjørtoft T, Bos AF, Einspieler C, Zhou Y, Schreiber MD, Marks JD, Drobyshevsky A.

AJNR Am J Neuroradiol. 2016 Oct 27. [Epub ahead of print]

BACKGROUND AND PURPOSE: Very preterm infants (birth weight, <1500 g) are at increased risk of cognitive and motor impairment, including cerebral palsy. These adverse neurodevelopmental outcomes are associated with white matter abnormalities on MR imaging at term-equivalent age. Cerebral palsy has been predicted by analysis of spontaneous movements in the infant termed "General Movement Assessment." The goal of this study was to determine the utility of General Movement Assessment in predicting adverse cognitive, language, and motor outcomes in very preterm infants and to identify brain imaging markers associated with both adverse outcomes and aberrant general movements. MATERIALS AND METHODS: In this prospective study of 47 preterm infants of 24-30 weeks' gestation, brain MR imaging was performed at term-equivalent age. Infants underwent T1- and T2-weighted imaging for volumetric analysis and DTI. General movements were assessed at 10-15 weeks' postterm age, and neurodevelopmental outcomes were evaluated at 2 years by using the Bayley Scales of Infant and Toddler Development III. RESULTS: Nine infants had aberrant general movements and were more likely to have adverse neurodevelopmental outcomes, compared with infants with normal movements. In infants with aberrant movements, Tract-Based Spatial Statistics analysis identified significantly lower fractional anisotropy in widespread white matter tracts, including the corpus callosum, inferior longitudinal and fronto-occipital fasciculi, internal capsule, and optic radiation. The subset of infants having both aberrant movements and abnormal neurodevelopmental outcomes in cognitive, language, and motor skills had significantly lower fractional anisotropy in specific brain regions. CONCLUSIONS: Aberrant general movements at 10-15 weeks' postterm are associated with adverse neurodevelopmental outcomes and specific white matter microstructure abnormalities for cognitive, language, and motor delays.

PMID: 27789448

26. Counting cases of cerebral palsy.

Platt MJ.

Dev Med Child Neurol. 2016 Oct 31. doi: 10.1111/dmcn.13304. [Epub ahead of print]

[This commentary is on the original article by Toyokawa et al]

PMID: 27797113

27. Significance of oligohydramnios in preterm small-for-gestational-age infants for outcome at 18 months of age.

Sasahara J, Ishii K, Umehara N, Oba M, Kiyoshi K, Murakoshi T, Tanemoto T, Ishikawa H, Ichizuka K, Yoshida A, Tanaka K, Ozawa K, Sago H.

J Obstet Gynaecol Res. 2016 Nov;42(11):1451-1456. doi: 10.1111/jog.13074. Epub 2016 Jun 29.

AIM: The aim of this study was to evaluate the association between oligohydramnios and other perinatal factors in preterm small-for-gestational-age (SGA) infants who had cerebral palsy at 18 months of age or who had died before this age. METHODS: This retrospective study included 320 infants with birthweights < 3rd percentile delivered between 22 and 33 complete weeks of gestation. We evaluated the incidence of CP at 18 months of age and of death before this age. The significant risk factors, including oligohydramnios, of CP or death of preterm SGA infants were evaluated by logistic regression analysis. RESULTS: The incidence of CP or death was 47/320 (14.7%), consisting of 24/320 (7.5%) cases of CP and 23/320 (7.2%) cases of death. Oligohydramnios (adjusted odds ratio, 2.18; 95% confidence interval, 1.07-4.45) and gestational age (adjusted odds ratio, 0.76; 95% confidence interval, 0.66-0.87) were independently correlated with outcome. CONCLUSION: The incidence of adverse outcomes was approximately 15% in preterm SGA infants. SGA infants born with oligohydramnios may be at increased risk for CP or death compared to those with normal amniotic volume.

PMID: 27352940