1. Effect of Core Stability Exercises on Hand Functions in Children With Hemiplegic Cerebral Palsy
Hanaa Mohsen Abd-Elfattah, Sobhy Mahmoud Aly

Objective: To investigate the effectiveness of core stability exercises on hand functions in children with hemiplegic cerebral palsy. Methods: Fifty-two children with hemiplegic cerebral palsy ranging in age from 6 years to 8 years were enrolled in this study. They were randomly assigned to two (control and study) groups. The control group received the selected physiotherapy exercises, and the study group received the same selected physiotherapy exercise program and core stability exercises. Time motor performance, gross manual dexterity, and upper extremity skills assessed using the Jebsen Taylor Hand Function Test, Box and Block Test, and Quality Upper Extremity Skill Test, respectively, were measured before and after 12 weeks of the treatment program. Results: There were significant improvements in both groups by comparing the mean values of all measured variables before and after treatment (p<0.05). There were significant differences between the control and study groups with respect to all measured variables when comparing the post-treatment outcomes (p<0.05). Conclusion: This study suggests that core stability exercises can be an effective intervention that may improve hand functions in children with hemiplegic cerebral palsy.

PMID: 33557483

2. Validity and reliability of the Selective Control of the Upper Extremity Scale in children with upper motor neuron lesions
Jan Lieber, Thomas Gartmann, Jeffrey W Keller, Hubertus J A van Hedel

Purpose: We evaluated the validity and intra-, inter-, and test-retest reliability of the Selective Control of the Upper Extremity Scale (SCUES) sum and item scores in patients with upper motor neuron lesions. Methods: Thirty-one boys and 15 girls (mean age ± SD: 11 years 1 month ± 3 years 9 month) with upper motor neuron lesions participated. We correlated SCUES scores with the range of motion items of the Melbourne Assessment 2 (MA2) and Box and Block Test (BBT) to establish concurrent validity and compared scores between the more and less affected side for discriminative validity. Intra-class correlation coefficients (ICC) and smallest detectable changes (SDC) indicated relative and absolute reliability. Results: For the more affected side, SCUES sum scores correlated well with MA2 (ρ = 0.83) and BBT (ρ = 0.77), and reliability proved high for intra-rater (ICC = 0.93, SDC = 2.55), inter-rater (ICC = 0.86, SDC = 3.58), and test-retest (ICC = 0.98, SDC = 1.41) reliability. Reliability of single items varied from 0.64 (inter-rater elbow) to 0.98 (intra-rater elbow). Limb and item scores were lower for the more affected side. Conclusion: The SCUES limb and item scores seem valid and reliable in children with upper motor neuron lesions. While future studies should evaluate the responsiveness of the SCUES, we recommend that the
same rater should score a patient twice. Implications for rehabilitation The SCUES assesses selective voluntary motor control and appears valid and reliable in patients with upper motor neuron lesions. Test-retest reliability of the SCUES seems excellent. SCUES single item scores show concurrent validity and acceptable reliability. Limb and item scores are significantly lower for the more affected side.

PMID: 33577357

3. Adaptive Working Memory Training Can Improve Executive Functioning and Visuo-Spatial Skills in Children With Pre-term Spastic Diplegia

Maria Chiara Di Lieto, Chiara Pecini, Paola Brovedani, Giuseppina Sgandurra, Marta Dell'Omo, Anna Maria Chilosi, Andrea Guzzetta, Silvia Perazza, Elisa Sicola, Giovanni Cioni


Pre-term spastic diplegia (pSD) due to periventricular leukomalacia is a form of cerebral palsy in which weaknesses in executive functions are reported beyond the core visuo-spatial deficits. The study aimed at improving executive functioning and visuo-spatial skills with an evidence-based training focused on working memory in children with pSD. The intervention study followed a stepped wedge design. 19 children with pSD (11 female and 8 male; age range: 4;1-13;1 years), mild to moderate upper limb impairment and Verbal Intelligence Quotient (VIQ) >80 participated to the study. The children were trained with a home-based adaptive working memory training (CogMed®) over a 5-week period. The primary outcome measure was the CogMed Improvement index; pre- and post-test explorative neuropsychological assessment was conducted with a subset of tests from the NEPSY-II battery. Working memory training in children with pSD significantly improved trained working memory abilities (CogMed indices) as well as non-trained skills, such as visuo-spatial skills, inhibition of automatic responses and phonological processing. The results suggest that standard rehabilitation schedules for children with pSD should be integrated with trainings on executive functions.

PMID: 33551960

4. Assessments and Interventions for Spasticity in Infants With or at High Risk for Cerebral Palsy: A Systematic Review

Lauren Ayala, Sarah Winter, Rachel Byrne, Darcy Fehlings, Alison Gehred, Lisa Letzkus, Garey Noritz, Madison C B Paton, Lindsay Pietruszewski, Nathan Rosenberg, Kelly Tanner, Jilda Vargus-Adams, Iona Novak, Nathalie L Maitre


Background: The majority of children with cerebral palsy develop spasticity, which interferes with motor development, function, and participation. This systematic review appraised current evidence regarding assessments and interventions for spasticity in children aged less than two years with or at high risk for cerebral palsy and integrated findings with parent preferences. Methods: Five databases (CINAHL, EMBASE, OVID/Medline, SCOPUS, and PsycINFO) were searched. Included articles were screened using PRISMA guidelines. Quality of the evidence was reviewed by two independent reviewers using Quality Assessment of Diagnostic Accuracy Studies, second edition (QUADAS-2), the RTI Item Bank on Risk of Bias and Precision of Observational Studies (RTI), or The Cochrane Collaboration's tool for assessing risk of bias in randomized trials (RoB). An online survey was conducted regarding parent preferences through social media channels. Results: Twelve articles met inclusion criteria. No high-quality assessment tool emerged for this population. Six interventions (botulinum toxin-A, orthotic use, radial extracorporeal shock wave therapy, erythropoietic stimulating agents, medical cannabis, and homeopathy) were identified. There was low-quality evidence for the use of botulinum toxin-A and radial extracorporeal shock wave therapy to improve short-term outcomes. Survey respondents indicated that spasticity assessments and interventions are highly valued, with nonpharmacologic interventions ranked most preferably. Conclusions: Further research is needed to validate assessments for spasticity in children younger than two years. Conditional recommendations can be made for botulinum toxin-A and radial extracorporeal shock wave therapy based on low level of evidence to reduce spasticity in children aged less than two years.

PMID: 33563492
5. Reconstructive hip surgery in children with cerebral palsy: factors influencing risk of femoral head avascular necrosis
Arya Minaie, Margaret Smythe, Joe Eric Gordon, Perry Schoenecker, Pooya Hosseinzadeh


Femoral head avascular necrosis (AVN) is a complication of hip reconstruction in children with cerebral palsy (CP) with uncertain incidence. The purpose of this study was to establish the rate of AVN in children undergoing hip reconstruction and to identify the factors associated with AVN. Medical records and radiographs of patients aged 1-18 with a diagnosis of CP who have undergone hip reconstruction over a 9-year period (2010-2018) with at least 1-year follow-up were reviewed. Age at time of surgery, sex, Gross Motor Function Classification System (GMFCS), procedure(s), preoperative migration percentage (MP), and signs of AVN on anterioposterior pelvis radiographs at routine follow-up visits (3 months, 6 months, and years 1-9, annually) according to the Bucholz-Ogden (BO) and Kalamchi-MacEwen (KM) classification systems were recorded. Univariate analysis was used to test for significance. Three hundred forty-nine hips in 209 patients met our inclusion criteria. Eleven hips (rate; 3%), in 11 patients, developed AVN. Classification of AVN was found to consist of: class I-IV (36%), class II-III [BO: 4 (36%) [KM: 3 (27%)], and Class IV- [BO: 2 (18%) [KM: 3 (27%)]. Average follow-up was of 3.5 years. The AVN cohort displayed older age (11.0 ± 2.8 vs. 8.1 ± 3.4; P = 0.005). Open reduction was associated with increased AVN, occurring in 9% of the hips (P = 0.004). No significant association was found between preoperative MP, GMFCS, or acetabular osteotomy. The method of open reduction (anterior vs. medial) was not significantly associated with AVN (P = 0.4471). In this large series, the observed rate of AVN was 3%, associated with open reductions at time of hip reconstruction. If possible, closed reduction would be preferred at time of hip reconstruction in children with CP to lower the risk of AVN. Level of evidence: III.

PMID: 33570364

6. Save the Hip-Hip Surveillance in Cerebral Palsy is the Need of the Hour!!
Ashok N Johari


PMID: 33569094

7. The Impact of Spinal Fusion on Hip Displacement in Cerebral Palsy
Mutlu Cobanoglu, Brian Po-Jung Chen, Lucio Perotti, Kenneth Rogers, Freeman Miller


Background and study aims: The aims of this study were to determine the risk of progressive hip subluxation in children with CP after spinal fusion for scoliosis and how frequent the hips follow-up should be scheduled. Patients and methods: Pelvis radiography [migration index (MI) and pelvic obliquity (PO)] of Gross Motor Function Classification System (GMFCS) levels IV and V children with CP who received spinal fusion and pelvic fixation were reviewed retrospectively. This population was categorized into three groups based on the MI at spinal fusion: G1 = 0-29%; G2 = 30-59%; and G3 = 60-100%. Results: Fifty children (age 7.5-15.0 years) and categorized into 3 groups (G1 = 19, G2 = 23, G3 = 8; 100 hips in total). Preoperative and last follow-up MI were 22 ± 7% and 30 ± 20% (G1), 41 ± 9% and 43 ± 22% (G2), 92 ± 15% and 97 ± 10% (G3). The MIs at spinal fusion between groups were statistically different (p < 0.001). In G1, the mean MI progression was 5% and 25% at 12 months and 62 months, respectively. In G2, the mean MI progression was 9% and 25% at 12 months and 32 months, respectively. The progression more than 10% occurred within 2 years in G1 and within 1 year in G2. There was no difference between groups based on preoperative degree of PO (p = 0.653) and correction rate in PO (p = 0.421). Conclusions: In GMFCS IV and V children with the highest risk for progression occurred with increasing preoperative MI, especially over 50%. Hips should be monitored continuously after spinal fusion until hip stability is documented.

PMID: 33569112
8. Hip Displacement in Cerebral Palsy: The Role of Surveillance
Alaric Aroojis, Nihit Mantri, Ashok N Johari


Introduction: Hip displacement is common in cerebral palsy (CP) and is related to the severity of neurological and functional impairment. It is a silent, but progressive disease, and can result in significant morbidity and decreased quality of life, if left untreated. The pathophysiology of hip displacement in CP is a combination of hip flexor-adductor muscle spasticity, abductor muscle weakness, and delayed weight-bearing, resulting in proximal femoral deformities and progressive acetabular dysplasia. Due to a lack of symptoms in the early stages of hip displacement, the diagnosis is easily missed. Awareness of this condition and regular surveillance by clinical examination and serial radiographs of the hips are the key to early diagnosis and treatment. Hip surveillance programmes: Several population-based studies from around the world have demonstrated that universal hip surveillance in children with CP allows early detection of hip displacement and appropriate early intervention, with a resultant decrease in painful dislocations. Global hip surveillance models are based upon the patients' age, functional level determined by the Gross Motor Function Classification system (GMFCS), gait classification, standardized clinical exam, and radiographic indices such as the migration percentage (MP), as critical indicators of progressive hip displacement. Conclusion: Despite 25 years of evidence showing the efficacy of established hip surveillance programmes, there is poor awareness among healthcare professionals in India about the importance of regular hip surveillance in children with CP. There is a need for professional organizations to develop evidence-based guidelines for hip surveillance which are relevant to the Indian context.

PMID: 33569095

9. Ankle contractures are frequent among children with cerebral palsy and associated with lower gross motor function and degree of spasticity
Laerke Hartvig Krarup, Pia Kjaer Kristensen, Louise Strand, Sofie Langbo Bredtoft, Inger Mechlenburg, Kirsten Nordbye-Nielsen


Aim: To estimate yearly prevalence of ankle contractures among children with cerebral palsy (CP). Moreover, to investigate if age, gross motor function or spasticity are associated with ankle contracture. Methods: We examined yearly prevalence of ankle contractures among 933 children based on data from a national clinical quality database from 2012 to 2019. We used the Gross Motor Function Classification System (GMFCS), the Modified Ashworth Scale (MAS) to assess gross motor function and spasticity in the plantar flexors. Ankle contracture was defined as dorsiflexion with an extended knee equal to or below 0 degrees. Associations between age, GMFCS, spasticity and ankle contractures were analyzed using multivariable regression and presented as odds ratios (OR) with 95% confidence intervals (95%CI). Results: The prevalence of ankle contracture was 32% and did not change with calendar year. GMFCS IV-V compared to I-III (40.6% vs. 28.9%, OR = 1.5 (95%CI: 1.07-2.11) and MAS 2-4 compared to 0 (44.6% vs. 24.4%, OR = 2.5 (95%CI: 1.59-3.91) were associated with a higher prevalence of ankle contracture. Age was not associated with ankle contracture. Conclusion: Ankle contractures are frequent among children with CP. Lower gross motor function and severe spasticity were associated with ankle contracture.

PMID: 33565134

10. Clinical utility of the over-ground bodyweight-supporting walking system Andago in children and youths with gait impairments
Hubertus J A van Hedel, Irene Rosselli, Sandra Baumgartner-Ricklin


Background: The Andago is a rehabilitation robot that allows training walking over-ground while providing bodyweight unloading (BWU). We investigated the practicability, acceptability, and appropriateness of the device in children with gait impairments undergoing neurorehabilitation. Concerning appropriateness, we investigated whether (i) stride-to-stride variability of the stride time and inter-joint coordination was higher when walking over-ground in Andago versus treadmill walking, and (ii) activation of antigravity leg muscles decreased with higher levels of BWU. Methods: Eighteen children and
adolescents with gait impairments participated in three sessions. Practicability was measured by determining the time needed to get a patient into and out of Andago, the accuracy of the BWU system, and other factors. Acceptability was measured by patients responding to questions, while six therapists evaluated the System Usability Scale. To determine appropriateness, the participants were equipped with surface electromyography (sEMG) electrodes, electrogoniometers, and accelerometers. Various parameters were compared between walking over-ground and on a treadmill, and between walking with three different levels of BWU (median: 20%, 35%, and 50% of the bodyweight) over-ground. Results: Practicability: the average time needed to get in and out of Andago amounted to 60 s and 16 s, respectively. The BWU system seemed accurate, especially at higher levels. We experienced no technical difficulties and Andago prevented 12 falls. However, participants had difficulties walking through a door without bumping into it. Acceptability: after the second session, nine participants felt safer walking in Andago compared to normal walking, 15 preferred walking in Andago compared to treadmill walking, and all wanted to train again with Andago. Therapists rated the usability of the Andago as excellent. Appropriateness: stride-to-stride variability of stride duration and inter-joint coordination was higher in Andago compared to treadmill walking. sEMG activity was not largely influenced by the levels of BWU investigated in this study, except for a reduced M. Gluteus Medius activity at the highest level of BWU tested. Conclusions: The Andago is a practical and well-accepted device to train walking over-ground with BWU in children and adolescents with gait impairments safely. The system allows individual stride-to-stride variability of temporospatial gait parameters without affecting antigravity muscle activity strongly. Trial registration: ClinicalTrials.gov Identifier: NCT03787199.

PMID: 33557834

11. Changes in the bony alignment of the foot after tendo-Achilles lengthening in patients with planovalgus deformity
Nak Tscheol Kim, Young Tae Lee, Moon Seok Park, Kyoungh Min Lee, Oh Sang Kwon, Ki Hyuk Sung


Background: This study was performed to investigate the change in the bony alignment of the foot after tendo-Achilles lengthening (TAL) and the factors that affect these changes in patients with planovalgus foot deformity. Methods: Consecutive 97 patients (150 feet; mean age 10 years; range 5.1-35.7) with Achilles tendon contracture (ATC) and planovalgus foot deformity who underwent TAL were included. All patients underwent preoperative and postoperative weight-bearing anteroposterior (AP) or lateral (LAT) foot radiographs. Changes in AP talo-1st metatarsal angle, AP talo-2nd metatarsal angle, LAT talo-1st metatarsal angle, and calcaneal pitch angle and the factors affecting such changes after TAL were analyzed using linear mixed model. Results: There were no significant change in AP talo-1st metatarsal angle and AP talo-2nd metatarsal angle after TAL in patients with cerebral palsy (CP) (p = 0.236 and 0.212). However, LAT talo-1st metatarsal angle and calcaneal pitch angle were significantly improved after TAL (13.0°, p < 0.001 and 4.5°, p < 0.001). Age was significantly associated with the change in LAT talo-1st metatarsal angle after TAL (p = 0.028). The changes in AP talo-1st metatarsal angle, AP talo-2nd metatarsal angle, and calcaneal pitch angle after TAL were not significantly associated with the diagnosis (p = 0.879, 0.903, and 0.056). However, patients with CP showed more improvement in LAT talo-1st metatarsal angle (- 5.0°, p = 0.034) than those with idiopathic cause. Conclusion: This study showed that TAL can improve the bony alignment of the foot in patients with planovalgus and ATC. We recommend that physicians should consider this study’s findings when planning operative treatment for such patients.

PMID: 33557891

12. The respiratory problems of patients with cerebral palsy requiring hospitalization: Reasons and solutions
Meltem Kürtül Çakar, Güzin Cinel


Objective: Recurrent respiratory problems are common in patients with cerebral palsy (CP). Detection of risk factors could prevent respiratory problems and reduce permanent lung damage. We aimed to provide a multidisciplinary approach for the causes, frequency, and concomitant problems and solutions of respiratory problems of these patients. Method: Medical records of CP patients who were followed-up and treated due to respiratory problems requiring hospitalization were examined retrospectively, and factors affecting the frequency and duration of hospitalization in wards and in the intensive care unit (ICU) were evaluated. Results: Among 292 hospitalizations of 83 CP patients, 91% of them were hospitalized for pneumonia, and only 15.4% of them had major aspiration-related pneumonia in the history. 97.4% of the hospitalized patients had swallowing dysfunction in detailed history, but videofluoroscopic swallowing studies could be performed only to 24 patients and aspiration...
had been demonstrated in 96%. Patients who had been hospitalized more than two times during the oral feeding period, before switching to artificial feeding (nasogastric tube, gastric tube +/- fundoplication), had higher total and ICU hospitalization frequency/duration than the patients who had been hospitalized twice or less (0.12 vs. 0.17/0.005106 vs. 0.005353).

Conclusion: The most common pulmonary complication in patients with CP is pneumonia due to chronic aspiration. To prevent recurrent lung infections and consequently chronic respiratory failure, it is necessary to evaluate these patients in terms of feeding difficulties in the early period with appropriate methods and apply interventions before affecting the lung parenchyma.

PMID: 33559955

13. Factors associated with oropharyngeal dysphagia diagnosed by videofluoroscopy in children with cerebral palsy [Article in English, Spanish]
N González-Rozo, J J Pérez-Molina, Y B Quíñones-Pacheco, L E Flores-Fong, A Rea-Rosas, J L Cabrales-deAnda


Introduction and aims: Oropharyngeal dysphagia (OD) occurs in children with cerebral palsy. It is important to investigate its relationship with some variables, and the objective of this study was to identify factors associated with OD. Materials and methods: Case-control study in patients with cerebral palsy from 8 months to 15 years of age, from November 2018 to November 2019, approved by the Ethics Committee. The diagnosis of OD was made by videofluoroscopy when there was nasopharyngeal reflux, stagnation in the vallecular sinuses, in the piriform sinuses, penetration, and aspiration. The independent variables were type of cerebral palsy, gross motor impairment classified into five levels, nutritional status and comorbidities. One case with OD was included and the next one without alterations in videofluoroscopy was control. The variables were compared with Chi square and Student's t. The association was measured with odds ratio. The confidence interval was 95%. Results: Thirty patients with OD and 30 without OD were studied. Sex, age, birth weight, and gestational age had a similar distribution in the two groups. From the data perceived by the mothers at the time of feeding, the greater frequency of the difficulty in the transfer of the food bolus in the group with OD showed a statistically significant difference (P<.001) and of the studied factors, the level V of the gross motor involvement was associated with a higher frequency of OD. Conclusions: OD was associated with level V of gross motor involvement.

PMID: 33573860

14. Traumatic dental injuries in special health care needs children and association with obesity
Sakeenabi Basha, Roshan Noor Mohamed, Yousef Al-Thomali, Amal Adnan Ashour, Fatma Salem Al Zahrani, Nada Eid Almutair


Background: Special needs children are at a higher risk of dental trauma because of neurological, physical, mental, and behavioral impairments. They are also at higher risk of developing obesity due to the side effects of medication. Objective: Assess the association between traumatic dental injuries (TDIs) and obesity in children with special health care needs. Design: Analytical cross-sectional study. Setting: Schools for special needs children. Study population and methods: Special needs children with a diagnosis of TDI according to the Andreasen criteria were included in the study. Data on the disability status were obtained from a national demographic survey in 2016. Demographic and dental variables were measured for analysis. Multivariable logistic regression was used to analyse any relationship between TDI prevalence and obesity. Main outcome measure: Relationship of body mass index (BMI) to TDI prevalence. Sample size: 350 (131 boys and 219 girls) special needs weight children (20.6%) (P=.035), but BMI category was not statistically significant in the regression analysis (P=.541), which showed that children with an overjet of >3 mm were 4.82 times (CI: 2.55-9.09, P=.001) more likely to have TDI than children with an overjet of ≤3 mm. Those with inadequate lip coverage were 2.85 times (CI: 1.49-5.44, P=.002) more likely to have TDI. Children with cerebral palsy were 3.18 times (CI: 1.89-11.32, P=.024) more likely to have TDI than children with other disabilities. Conclusion: The study showed a significant association between TDI prevalence and increased overjet, inadequate lip coverage, and cerebral palsy. The prevalence of TDI among obese special needs children was statistically significant according to bivariate analysis, but not in a multivariate analysis that adjusted for other variables. Limitations: Causal relationship cannot be established with cross-sectional study.
15. Adults with cerebral palsy require ongoing neurologic care: A systematic review
Sarah E Smith, Mary Gannotti, Edward A Hurvitz, Frances E Jensen, Linda E Krach, Michael C Krue, Michael E Msall, Garey Noritz, Deepa S Rajan, Bhooma R Aravamuthan


Cerebral palsy (CP) neurologic care and research efforts typically focus on children. However, most people with CP are adults. Adults with CP are at increased risk of new neurologic conditions, like stroke and myelopathy, that require ongoing neurologic surveillance to distinguish them from baseline motor impairments. Neurologic factors could also contribute to the motor function decline, chronic pain, and chronic fatigue that are commonly experienced by adults with CP. Based on a systematic literature review, we suggest: 1) guidelines for neurologic surveillance and neurologist referral; and 2) clinical research questions regarding the evolving neurologic risks for adults with CP. This article is protected by copyright. All rights reserved.

PMID: 33550911

16. Ultrasound shearwave elastography to characterize muscles of healthy and cerebral palsy children
Pauline Lallemant-Dudek, Claudio Vergari, Guillaume Dubois, Véronique Forin, Raphaël Vialle, Wafa Skalli


Shear wave elastography (SWE) is an ultrasound technique to obtain soft tissue mechanical properties. The aim of this study was to establish the reliability of SWE in young children, define reference data on healthy ones and compare the shear modulus of healthy and spastic muscles from cerebral palsy (CP). The reproducibility is evaluated: at rest, on 7 children without any musculoskeletal pathology by 3 different operators, on 2 muscles: biceps brachii long head and medial gastrocnemius. The comparison study was made, on the same 2 muscles, at rest and under passive stretching, with a control group (29 healthy children), a spastic group (spastic muscles of 16 children from CP) and a non-spastic group (non-spastic muscles of 14 children from CP). The intra-operator reliability and inter-operator reliability, in terms of standard deviation, were 0.6 kPa (11.2% coefficient of variation (CV)) and 0.8 kPa (14.9% CV) for the biceps, respectively, and 0.4 kPa (11.5% CV) and 0.5 kPa (13.8% CV) for the gastrocnemius. At rest, no significant difference was found. Under passive stretching, the non-spastic CP biceps were significantly stiffer than the control ones (p = 0.033). Spastic gastrocnemius had a higher shear modulus than in the control muscles (p = 0.0003) or the non-spastic CP muscles (p = 0.017). CP stretched medial gastrocnemius presented an abnormally high shear moduli for 50% of patients.

PMID: 33550625

17. Stakeholder consensus for decision making in eye-gaze control technology for children, adolescents and adults with cerebral palsy service provision: findings from a Delphi study
Petra Karlsson, Tom Griffiths, Michael T Clarke, Elegast Mombaliu, Kate Himmelmann, Saranda Bekteshi, Abigail Allsop, René Pereksles, Claire Galea, Margaret Wallen


Background: Limited research exists to guide clinical decisions about trialling, selecting, implementing and evaluating eye-gaze control technology. This paper reports on the outcomes of a Delphi study that was conducted to build international stakeholder consensus to inform decision making about trialling and implementing eye-gaze control technology with people with cerebral palsy. Methods: A three-round online Delphi survey was conducted. In Round 1, 126 stakeholders responded to questions identified through an international stakeholder Advisory Panel and systematic reviews. In Round 2, 63 respondents rated the importance of 200 statements generated by in Round 1. In Round 3, 41 respondents rated the importance of the 105 highest ranked statements retained from Round 2. Results: Stakeholders achieved consensus on 94 of the original 200 statements. These statements related to person factors, support networks, the environment, and technical aspects to consider
during assessment, trial, implementation and follow-up. Findings reinforced the importance of an individualised approach and that information gathered from the user, their support network and professionals are central when measuring outcomes. Information required to support an application for funding was obtained. Conclusion: This Delphi study has identified issues which are unique to eye-gaze control technology and will enhance its implementation with people with cerebral palsy.

PMID: 33568101

18. The future of General Movement Assessment: The role of computer vision and machine learning - A scoping review
Nelson Silva, Dajie Zhang, Tomas Kulvicius, Alexander Gail, Carla Barreiros, Stefanie Lindstaedt, Marc Kraft, Sven Bölte, Luise Poustka, Karin Nielsen-Saines, Florentin Wörgötter, Christa Einspieler, Peter B Marschik


Background: The clinical and scientific value of Prechtl general movement assessment (GMA) has been increasingly recognised, which has extended beyond the detection of cerebral palsy throughout the years. With advancing computer science, a surging interest in developing automated GMA emerges. Aims: In this scoping review, we focused on video-based approaches, since it remains authentic to the non-intrusive principle of the classic GMA. Specifically, we aimed to provide an overview of recent video-based approaches targeting GMs; identify their techniques for movement detection and classification; examine if the technological solutions conform to the fundamental concepts of GMA; and discuss the challenges of developing automated GMA. Methods and procedures: We performed a systematic search for computer vision-based studies on GMs. Outcomes and results: We identified 40 peer-reviewed articles, most (n = 30) were published between 2017 and 2020. A wide variety of sensing, tracking, detection, and classification tools for computer vision-based GMA were found. Only a small portion of these studies applied deep learning approaches. A comprehensive comparison between data acquisition and sensing setups across the reviewed studies, highlighting limitations and advantages of each modality in performing automated GMA is provided. Conclusions and implications: A "method-of-choice" for automated GMA does not exist. Besides creating large datasets, understanding the fundamental concepts and prerequisites of GMA is necessary for developing automated solutions. Future research shall look beyond the narrow field of detecting cerebral palsy and open up to the full potential of applying GMA to enable an even broader application.

PMID: 33571849

Hortensia Gimeno, Helene J Polatajko, Jean-Pierre Lin, Victoria Cornelius, Richard G Brown


Objective: To explore preliminary effectiveness of the Cognitive Orientation to daily Occupational Performance (CO-OP) Approach in improving outcomes in childhood-onset hyperkinetic movement disorders (HMDs) including dyskinetic cerebral palsy following deep brain stimulation (DBS) across UK clinical occupational therapists. Methods: Randomized, multiple-baseline, Single Case Experimental Design N-of-1 trial with replications across participants. Five self-selected goals were identified: three goals were worked on during CO-OP and two goals were left untreated and used to assess skills transfer. Participants were between 6 and 21 years and had received DBS surgery with baseline Manual Ability Classification System (MACS) levels I-IV. Participants were randomized to typical or extended baseline (2 vs. 6 weeks), followed by 10 weekly individual CO-OP sessions. The primary outcome was functional performance measured by the Performance Quality Rating Scale-Individualized (PQRS-I), assessed before, during, and following treatment. Outcome assessors were blinded to baseline allocation, session number, and assessment time. A non-overlapping index, Tau-U, was used to measure effect size. Results: Of the 12 participants recruited, 10 commenced and completed treatment. In total, 63% of trained goals improved with effect sizes 0.66-1.00 ("moderate" to "large" effect), seen for all children in at least one goal. Skills transfer was found in 37% of the untrained goals in six participants. Conclusions: Cognitive strategy use improved participant-selected functional goals in childhood-onset HMD, more than just practice during baseline. Preliminary effectiveness is shown when the intervention is delivered in clinical practice by different therapists in routine clinical settings.

PMID: 33553070
20. Consensus of physician behaviours to target for early diagnosis of cerebral palsy: A Delphi study
Lynda McNamara, Karen M Scott, Roslyn N Boyd, Iona Novak


Aims: Historically, the diagnosis of cerebral palsy has been made after 12 months of age, delaying access to crucial early intervention that optimises functional outcomes. This study aimed to identify and specify priority physician diagnostic behaviours to target in implementation interventions to increase the rate of diagnosis of cerebral palsy under 6 months of age in Australia. Methods: We conducted a two-round online Delphi study with a multi-professional expert panel of cerebral palsy researchers and clinicians. A reference group identified a six-item list of potential diagnostic behaviours, which were modifiable at the individual level, that could lead to an early cerebral palsy diagnosis. In the first survey, participants rated the importance of each item on a 10-point Likert scale and supplied their reasoning for this, and were able to suggest new behaviours. In the second survey, participants ranked items in order of priority. Results: All six items reached consensus for inclusion (100%). No new items were added to the list. Ranking identified the top three priorities for online physician implementation interventions: (i) refer for or conduct the General Movements Assessment; (ii) refer for or conduct the Hammersmith Infant Neurological Examination; and (iii) communication of the diagnosis. Conclusion: An online Delphi method can effectively inform tailored implementation intervention development. A consensus was achieved on the priority physician diagnostic behaviours to target in interventions to lower the age of cerebral palsy diagnosis in Australia.

PMID: 33556996

21. Movements and posture in infants born extremely preterm in comparison to term-born controls
Maria Örtqvist, Christa Einspieler, Peter B Marschik, Ulrika Ådén


Background: Identifying altered motor development at an early stage is crucial for infants born extremely preterm (EPT), as they face a high risk of long-term neurodevelopmental impairment. The Prechtl General Movement Assessment (GMA), including the Motor Optimality Score Revised (MOS-R), can provide important insights into these infants’ later neurodevelopmental function. Aims: To compare age-specific movements and postures in infants born EPT compared to term-born controls at three months corrected age. Study design: A retrospective observational study design. Subjects: 53 infants born EPT (mean gestational age 25 weeks; 23-26) were included and matched for gender and recording age with 53 term-born controls (mean gestational age 40 weeks, 37-41). Outcome measures: GMA including the MOS-R at three months corrected age (re-analysis of video-recordings). Results: Of the infants born EPT, 19% showed aberrant fidgety movements (FMs); all term-born infants had normal FMs. There was a significant difference in MOS-R (p≤0.001) between controls (median = 26, IQR 26-28) and EPT infants (median = 18, IQR 17-21), as well as in all subcategories of the MOS-R. The EPT group had a significantly higher number of infants showing atypical movement and postural patterns as well as a reduced repertoire for the age compared to the controls. All infants born EPT moved monotonously and jerky. P-values were all <0.001. Conclusion: Infants born EPT have an altered early motor development. The MOS-R may contribute to further understanding of motor performance in this group of children since it can detect neurological- and motor alterations at a very early age.

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22. Ectopic pregnancy: perinatal outcomes of future gestations and long-term neurological morbidity of the offspring
Melanie Shanie Roitman, Tamar Wainstock, Eyal Sheiner, Tom Leibson, Gali Pariente


Purpose: To evaluate perinatal outcomes and long-term neurological morbidity of offspring to mothers with a history of ectopic pregnancy. Methods: In this retrospective study, perinatal outcomes and long-term neurological morbidity of offspring were assessed among mothers with a history of ectopic pregnancy, either medically or surgically treated. The study groups were followed until 18 years of age for neurological-related morbidity. For perinatal outcomes, generalized estimated equation (GEE) models were used to control for confounders. A Kaplan-Meier survival curve was used to compare cumulative neurological morbidity incidence and Cox proportional hazards model was conducted to control for confounders. Results: A
total of 243,682 mothers were included; 1424 mothers (0.58%) had a previous ectopic pregnancy, of which 25.6% (n = 365) were treated medically, and 74.3% (n = 1059) were treated surgically. Using GEE models, controlling for confounders, both surgically and medically treated ectopic pregnancies were noted as independent risk factors for preterm delivery in the subsequent pregnancies. Maternal history of surgically treated ectopic pregnancy was also independently associated with cesarean delivery. Offspring to mothers with previous ectopic pregnancy had comparable rates of long-term neurological morbidity. In the Cox proportional hazards model, controlling for confounders, being born to a mother with a history of previous ectopic pregnancy was not found to be independently associated with long-term neurological morbidity of offspring. Conclusions: Maternal history of ectopic pregnancy is independently associated with preterm delivery. However, offspring of mothers with a history of ectopic pregnancy are not at an increased risk for long-term neurological morbidity.

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23. COHESION: core outcomes in neonatal encephalopathy (protocol)
Fiona A Quirke, Patricia Healy, Elaine Ni Bhraonain, Mandy Daly, Linda Biesty, Tim Hurley, Karen Walker, Shireen Meher, David M Haas, Frank H Bloomfield, Jamie J Kirkham, Eleanor J Molloy, Declan Devane


Background: Neonatal encephalopathy is a complex syndrome in infants that predominantly affects the brain and other organs. The leading cause is a lack of oxygen in the blood reaching the brain. Neonatal encephalopathy can result in mortality or complications later in life, including seizures, movement disorders and cerebral palsy. Treatment options for neonatal encephalopathy are limited mainly to therapeutic hypothermia, although other potential treatments are emerging. However, evaluations of the effectiveness of treatments are challenging because of heterogeneity and inconsistency in outcomes measured and reported between trials. In this paper, we detail how we will develop a core outcome set to standardise outcomes measured and reported upon for interventions for the treatment of neonatal encephalopathy. Methods: We will systematically review the literature to identify outcomes reported previously in randomised trials and systematic reviews of randomised trials. We will identify outcomes important to parents or caregivers of infants diagnosed with and who have received treatment for neonatal encephalopathy. We will do this by conducting in person or by video teleconferencing interviews with parents or caregivers in high- to middle-income countries. Stakeholders with expertise in neonatal encephalopathy (parents/caregivers, healthcare providers and researchers) will rate the importance of identified outcomes in an online Delphi survey using either a three-round Delphi survey or a "Real-Time" Delphi survey to which stakeholders will be allocated at random. Consensus meetings will take place by video conference to allow for an international group of stakeholder representatives to discuss and vote on the outcomes to include in the final core outcome set (COS). Discussion: More research is needed on treatments for neonatal encephalopathy. Standardising outcomes measured and reported in evaluations of the effectiveness of interventions for the treatment of neonatal encephalopathy will improve evidence synthesis and improve results reported in systematic reviews and meta-analysis in this area. Overall, this COS will allow for improved treatments to be identified, heterogeneity in research to be reduced, and overall patient care to be enhanced. Trial registration: This study is registered in the Core Outcome Measures for Effectiveness (COMET) database http://www.comet-initiative.org/Studies/Details/1270.

PMID: 33557892

24. Categorization of cerebral palsy cases: a different perspective
Mark I Evans, David W Britt


PMID: 33567328

25. REPLY TO "CATEGORIZATION OF CEREBRAL PALSY CASES: A DIFFERENT PERSPECTIVE"
Masahiro Nakao, Mitsutoshi Iwashita, Tomoaki Ikeda

26. The Emperor Has No Clothes: Evidence-Based Medicine for Cerebral Palsy
E Steve Roach


PMID: 33567330

27. A dynamic era in cerebral palsy research
No authors listed


PMID: 33556745

28. Insights From Genetic Studies of Cerebral Palsy
Sara A Lewis, Sheetal Shetty, Bryce A Wilson, Aris J Huang, Sheng Chih Jin, Hayley Smithers-Sheedy, Michael C Fahey, Michael C Kruer


Cohort-based whole exome and whole genome sequencing and copy number variant (CNV) studies have identified genetic etiologies for a sizable proportion of patients with cerebral palsy (CP). These findings indicate that genetic mutations collectively comprise an important cause of CP. We review findings in CP genomics and propose criteria for CP-associated genes at the level of gene discovery, research study, and clinical application. We review the published literature and report 18 genes and 5 CNVs from genomics studies with strong evidence of for the pathophysiology of CP. CP-associated genes often disrupt early brain developmental programming or predispose individuals to known environmental risk factors. We discuss the overlap of CP-associated genes with other neurodevelopmental disorders and related movement disorders. We revisit diagnostic criteria for CP and discuss how identification of genetic etiologies does not preclude CP as an appropriate diagnosis. The identification of genetic etiologies improves our understanding of the neurobiology of CP, providing opportunities to study CP pathogenesis and develop mechanism-based interventions.

PMID: 33551980

29. Pathogenic convergence of CNVs in genes functionally associated to a severe neuromotor developmental delay syndrome
Juan L García-Hernández, Luis A Corchete, Íñigo Marcos-Alcalde, Paulino Gómez-Puertas, Carmen Fons, Pedro A Lazo


Background: Complex developmental encephalopathy syndromes might be the consequence of unknown genetic alterations that are likely to contribute to the full neurological phenotype as a consequence of pathogenic gene combinations. Methods: To identify the additional genetic contribution to the neurological phenotype, we studied as a test case a boy, with a KCNQ2 exon-7 partial duplication, by single-nucleotide polymorphism (SNP) microarray to detect copy-number variations (CNVs). Results: The proband presented a cerebral palsy like syndrome with a severe motor and developmental encephalopathy. The SNP array analysis detected in the proband several de novo CNVs, nine partial gene losses (LRRC55, PCDH9, NALCN, RYR3, ELAVL2, CDH13, ATP1A2, SLC17A5, ANO3), and two partial gene duplications (PCDH19, EFNA5). The biological
functions of these genes are associated with ion channels such as calcium, chloride, sodium, and potassium with several membrane proteins implicated in neural cell-cell interactions, synaptic transmission, and axon guidance. Pathogenically, these functions can be associated to cerebral palsy, seizures, dystonia, epileptic crisis, and motor neuron dysfunction, all present in the patient. Conclusions: Severe motor and developmental encephalopathy syndromes of unknown origin can be the result of a phenotypic convergence by combination of several genetic alterations in genes whose physiological function contributes to the neurological pathogenic mechanism.

PMID: 33557955

30. 'Imaging and genetic information link aetiological mechanisms, structural brain injury patterns and functional outcomes in dyskinetic cerebral palsy in Osaka.'
No authors listed


PMID: 33556747

31. Cytokine and chemokine responses to injury and treatment in a nonhuman primate model of hypoxic-ischemic encephalopathy treated with hypothermia and erythropoietin


Predicting long-term outcome in infants with hypoxic-ischemic encephalopathy (HIE) remains an ongoing clinical challenge. We investigated plasma biomarkers and their association with 6-month outcomes in a nonhuman primate model of HIE with or without therapeutic hypothermia (TH) and erythropoietin (Epo). Twenty-nine Macaca nemestrina were randomized to control cesarean section (n = 7) or 20 min of umbilical cord occlusion (UCO, n = 22) with either no treatment (n = 11) or TH/Epo (n = 11). Initial injury severity was scored using 30-min arterial pH, base deficit, and 10-min Apgar score. Twenty-four plasma cytokines, chemokines, and growth factors were measured 3, 6, 24, 72, and 96 h after UCO. Interleukin 17 (IL-17) and macrophage-derived chemokine (MDC) differentiated the normal/mild from moderate/severe injury groups. Treatment with TH/Epo was associated with increased monocyte chemotactic protein-4 (MCP-4) at 3 h-6 h, and significantly lower MCP-4 and MDC at 24 h-72 h, respectively. IL-12p40 was lower at 24 h-72 h in animals with death/cerebral palsy (CP) compared to survivors without CP. Baseline injury severity was the single best predictor of death/CP, and predictions did not improve with the addition of biomarker data. Circulating chemokines associated with the peripheral monocyte cell lineage are associated with severity of injury and response to therapy, but do not improve ability to predict outcomes.

PMID: 33554708

Prevention and Cure

32. Preventing Brain Injury in the Preterm Infant-Current Controversies and Potential Therapies
Nathanael Yates, Alistair J Gunn, Laura Bennet, Simerdeep K Dhillon, Joanne O Davidson


Preterm birth is associated with a high risk of morbidity and mortality including brain damage and cerebral palsy. The development of brain injury in the preterm infant may be influenced by many factors including perinatal asphyxia, infection/inflammation, chronic hypoxia and exposure to treatments such as mechanical ventilation and corticosteroids. There are
currently very limited treatment options available. In clinical trials, magnesium sulfate has been associated with a small, significant reduction in the risk of cerebral palsy and gross motor dysfunction in early childhood but no effect on the combined outcome of death or disability, and longer-term follow up to date has not shown improved neurological outcomes in school-age children. Recombinant erythropoietin has shown neuroprotective potential in preclinical studies but two large randomized trials, in extremely preterm infants, of treatment started within 24 or 48 h of birth showed no effect on the risk of severe neurodevelopmental impairment or death at 2 years of age. Preclinical studies have highlighted a number of promising neuroprotective treatments, such as therapeutic hypothermia, melatonin, human amnion epithelial cells, umbilical cord blood and vitamin D supplementation, which may be useful at reducing brain damage in preterm infants. Moreover, refinements of clinical care of preterm infants have the potential to influence later neurological outcomes, including the administration of antenatal and postnatal corticosteroids and more accurate identification and targeted treatment of seizures.

PMID: 33562339

33. Umbilical cord blood CD34 + cells administration improved neurobehavioral status and alleviated brain injury in a mouse model of cerebral palsy
Yanqun Chang, Shouheng Lin, Yongsheng Li, Song Liu, Tianbao Ma, Wei Wei


Purpose: Cerebral palsy (CP) is the most common neuromuscular disease in children, and currently, there is no cure. Several studies have reported the benefits of umbilical cord blood (UCB) cell treatment for CP. However, these studies either examined the effects of UCB cell fraction with a short experimental period or used neonatal rat models for a long-term study which displayed an insufficient immunological reaction and clearance of human stem cells. Here, we developed a CP model by hypoxia-ischemic injury (HI) using immunodeficient mice and examined the effects of human UCB CD34+ hematopoietic stem cells (HSCs) on CP therapy over a period of 8 weeks. Methods: Sixty postnatal day-9 (P9) mouse pups were randomly divided into 4 groups (n = 15/group) as follows: (1) sham operation (control group), (2) HI-induced CP model, (3) CP model with CD34+ HSC transplantation, and (4) CP model with CD34- cell transplantation. Eight weeks after insult, the sensorimotor performance was analyzed by rotarod treadmill, gait dynamic, and open field assays. The pathological changes in brain tissue of mice were determined by HE staining, Nissl staining, and MBP immunohistochemistry of the hippocampus in the mice. Results: HI brain injury in mice pups resulted in significant behavioral deficits and loss of neurons. Both CD34+ HSCs and CD34- cells improved the neurobehavioral statuses and alleviated the pathological brain injury. In comparison with CD34- cells, the CD34+ HSC compartments were more effective. Conclusion: These findings indicate that CD34+ HSC transplantation was neuroprotective in neonatal mice and could be an effective therapy for CP.

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