

# Cerebral palsy research news

Monday 10 January 2022

**Cerebral Palsy Alliance** is delighted to bring you this free weekly bulletin of the latest published research into cerebral palsy. Our organisation is committed to supporting cerebral palsy research worldwide - through information, education, collaboration and funding. Find out more at <u>cerebralpalsy.org.au/our-research</u>

**Professor Nadia Badawi AM**CP Alliance Chair of Cerebral Palsy Research

Subscribe to CP Research News

#### **Interventions and Management**

#### 1. A Novel and Clinically Feasible Instrument for Quantifying Upper Limb Muscle Tone and Motor Function via Indirect Measure Methods

Chieh-Hsiang Hsu, Yu-Chen Lin, Hsiu-Yun Hsu, Hsiao-Feng Chieh, Chien-Ju Lin, Shih-Fu Ling, Fong-Chin Su, Li-Chieh Kuo

IEEE J Transl Eng Health Med. 2021 Dec 20;10:2100208. doi: 10.1109/JTEHM.2021.3136754. eCollection 2022.

Objective: Quantifying muscle tone is often based on a tester's subjective judgment in clinical settings. There is, however, a lack of suitable tools that can be used to objectively assess muscle tone. This study thus introduces a reliable, clinically-feasible device, called the Arm Circumference Motor Evaluation System (ACMES), for quantifying the muscle tone of upper limbs without using mechanical torque transducers. Methods: While the ACMES conducts continuously passive arm circumduction motions, the voltage and current of the driving motor is transduced into torque values via a least square approximation. A torque sensor and springs with different spring constants were used for the validity and reliability test in the first part of this study. Fifteen healthy adults and two patients who had experienced a stroke participated in the second part, which was a clinical experiment used to examine the in-vivo test-retest reliability and to explore the inspection differences between healthy and patient participants. Results: The results showed that the ACMES has high validity (R2: ~0.99) and reliability (R2: 0.96~0.99). The reliability of the ACMES used on human subjects was acceptable (R2: 0.83~0.85). The various muscle tone patterns could be found among healthy and stroke subjects via the ACMES. Conclusion: Clinically, abnormal muscle tone, which seriously affects motion performance, will be found in many diagnoses, such as stroke or cerebral palsy. However, objectively and feasibly measuring abnormal tone in modern clinical settings is still a challenging task. Thus, the ACMES was developed and tested to verify its feasibility as a measurement system for detecting the mechanical torque associated with muscle tone.

PMID: 34984110

## 2. Effect of upper extremity constraints on functional and dynamic postural control in children with hemiplegic cerebral palsy

Meysam Roostaei, Parvin Raji, Khosro Khademi Kalantari, Elham Faghihzadeh, Maria Fragala-Pinkham

Dev Neurorehabil. 2021 Dec 31;1-8. doi: 10.1080/17518423.2021.2020351. Online ahead of print.

Purpose: To compare the effects of upper extremity constraints on functional and dynamic postural control in children with hemiplegic cerebral palsy. Materials and methods: Twelve children with hemiplegic cerebral palsy and 12 typically developing children ages 5 to 12 years (GMFCS I-II) were evaluated with the Timed Up and Go (TUG) and Biodex Balance System during three upper extremity (UE) conditions: 1) Free UEs (no constraints), 2) Simple UE constraint (unaffected/

dominant UE constrained with a sling), and 3) Difficult UE constraint (Simple constraint plus the other UE holding cup of water). Results: The UE condition had significant effects on Overall Stability Index (OSI) (F(2,44) = 24.899, p < .001), Medial-Lateral Stability Index (MLSI) (F(2,44) = 4.380, p = .018), Anterior-Posterior Stability Index (F(2,44) = 6.187, p = .004), and TUG scores (F(2,44) = 113.372, p < .001). Group was significant for OSI (F(1,22) = 7.906, p = .010), MLSI (F(1,22) = 13.113, p = .002), and TUG (F(1,22) = 36.282, p < .001). Conclusions: The upper extremity appears to have a role in maintaining functional balance and postural stability in children with hemiplegic cerebral palsy and should be considered during intervention programs.

PMID: 34971522

#### 3. The unstable hip in children with cerebral palsy: does an acetabuloplasty add midterm stability? Matthias W Axt, Danielle L Wadley

J Child Orthop. 2021 Dec 1;15(6):564-570. doi: 10.1302/1863-2548.15.210154.

Purpose: This study addresses whether an additional pelvic procedure is superior to a varus derotation osteotomy femur (VDRO) alone in unstable hips in children with cerebral palsy (CP). Methods: All patients had unstable hips utilising the Melbourne Cerebral Palsy Hip Classification System (MCPHCS). We compared one group that underwent VDRO alone with one that had a combination of VDRO and Dega osteotomy (VDRO+). Measurements were taken before surgery, postoperatively, two years after surgery and at latest follow-up. Generalised estimating equations were used to account for known and unknown correlations between hips from bilateral cases. Results: In total, 74 hips in 57 children fulfilled the inclusion criteria. There was no outcome difference between Gross Motor Function Classification System levels III, IV and V. Age at time of operation ranged from three to 16 years (mean 9.8 years). Mean follow-up was 49.1 months. In the VDRO group (28 hips) migration percentage (MP) changed from 61% preoperative to a final value of 35.7%. In the VDRO+ group (46 hips) the MP changed from 64.4% to 19.3%. At final follow-up 15 hips (54%) were stable in the VDRO group, 37 hips (83%) in the VDRO+ group. The odds ratio (OR) of hip stability at final follow-up was 3.5-times higher in the VDRO+ group versus the VDRO group (OR = 3.9; 95% confidence interval = 1.5 to 9.7; p = 0.004). Conclusion: Reconstruction of unstable hips via VDRO + Dega in children with CP provides a higher likelihood of long-term stability than an isolated VDRO. Level of evidence: Level III, retrospective comparative study.

PMID: 34987666

#### 4. Percutaneous hamstring lengthening in cerebral palsy and the risk of neurovascular transection Thanase Ariyawatkul, Pathom Halilamien, Suwimon Tangwiwat, Busara Sirivanasandha, Pawinee Pangthipampai, Chatupon

Chotigavanichaya, Jidapa Wongcharoenwatana, Perajit Eamsobhana

J Ultrasound. 2022 Jan 7. doi: 10.1007/s40477-021-00620-9. Online ahead of print.

Purpose: Percutaneous hamstring lengthening is increasingly popular due to its simplicity, fast recovery rate, and low morbidity. Neurovascular anatomy changes due to knee flexion contracture and the precise proximity of peroneal nerve and Biceps femoris tendon are not well established. This study examined (1) the coronal distance between the peroneal nerve and lateral hamstring tendon ("PLD"), and (2) the distance between the popliteal vessels and medial hamstring tendons ("VMD") to determine the safe distance for percutaneous hamstring lengthening. Methods: This prospective study recruited cerebral palsy patients aged under 15 who needed hamstring lengthening. Ultrasonography was performed after the patients were anesthetized. PLDs and VMDs at popliteal angles (PAs) of 40°, 60°, and 80° knee flexions were collected. Results: Sixteen patients (32 knees) were enrolled. The mean minimum PLDs at PAs of 40°, 60°, and 80° were 3.5, 4.1, and 3.1 mm, respectively. The peroneal nerve physically touched the lateral hamstring tendon in 5/32 knees (15.6%). The mean minimum VMDs at PAs of 40°, 60°, and 80° were 19, 18.3, and 16.4 mm, respectively. One spastic diplegic patient had a minimum VMD < 3 mm on both sides. Changing the PAs demonstrated no statistical significance for both PLD and VMD (P value = 0.105 and 0.779, respectively). Conclusions: Percutaneous medial hamstring lengthening should be done with caution. We recommend open biceps femoris surgery, with preoperative ultrasonography (to check the PLD) or peroneal nerve palpation to reduce the risk of peroneal nerve transection.

## 5. Relation of the Sural Nerve and Medial Neurovascular Bundle With the Achilles Tendon in Children With Cerebral Palsy Treated by Percutaneous Achilles Tendon Lengthening

Ozan A Erdal, Baris Gorgun, Necip S Yontar, Ali E Terzibaşioğlu, Ilker A Sarikaya, Muharrem Inan

J Pediatr Orthop. 2022 Feb 1;42(2):e201-e205. doi: 10.1097/BPO.000000000002020.

Background: One of the most common treatment options for a short Achilles tendon (AT) in cerebral palsy is percutaneous AT lengthening using 3 hemisections. Because of proximity of neurovascular structures around the tendon, iatrogenic injury to them have been a concern about this technique. The sural nerve (SN) is under risk of injury at the site of the lateral incomplete cut, especially if it is done proximally. The medial neurovascular bundle is under injury risk at medial cuts. The aim of the article was to study the anatomical relations of the SN and medial neurovascular bundle to the AT, and define dangerous levels for injury with the help of magnetic resonance imaging (MRI). Methods: Patients operated for percutaneous Achilles lengthening were called for MRI investigation of the SN and medial neruvascular bundle integrity and their anatomical relation with the AT. The distance of 5 mm was taken as the threshold for increased risk of injury. Measurements were done on MRI at each cm from the insertion of the tendon on both medial and lateral sides, and at the level of the middle cut. Results: Thirty ankles operated and followed at least 1 year were included to the study. On the medial side, the tibial nerve, and the posterior tibial artery lied more than 5 mm away from the tendon at all levels in all patient. On the lateral side, the first 4 cm were relatively safe for the middle lateral cut, while increased risk of SN damage was detected in more proximal levels. Overall, 6 of 30 ankles had radiographically detectable SN injury. Conclusions: The first 4 cm of the AT on the lateral side was detected to be safe for the middle lateral directed cut, while whole tendon length were found to be safe for the first and the third cuts of the percutaneous Achilles lengthening surgery using 3 hemisections in children with cerebral palsy. Level of evidence: Level III.

PMID: 34995262

## 6. Reliability and validity of the Turkish version of the Selective Control Assessment of the Lower Extremity (SCALE) in children with spastic cerebral palsy

Merve Tunçdemir, Sefa Üneş, Jale Karakaya, Mintaze Kerem Günel

Disabil Rehabil. 2022 Jan 7;1-5. doi: 10.1080/09638288.2021.2022783. Online ahead of print.

Purpose: This study aims to translate the Selective Control Assessment of the Lower Extremity (SCALE) into Turkish language, assess its reliability and validity in children with spastic cerebral palsy. Materials and methods: Fifty-two children with CP (mean age 9 years 8 months, range 4-18 years) included in this cross-sectional study. Intra- and interrater reliability was assessed by intraclass correlation coefficient (ICC). The SCALE was correlated with the Gross Motor Function Classification System (GMFCS), the Physician's Rating Scale (PRS), and Gross Motor Function Measurement (GMFM) to assess validity. Results: Intra- and interrater reliability of the SCALE were excellent (ICC > 0.75). SCALE and GMFCS (r = 0.786, p < 0.001), SCALE and PRS (r = 0.761, p < 0.001), SCALE and GMFM (r = 0.863, p < 0.001) were highly correlated. SCALE scores differed significantly between GMFCS levels and between types of spastic CP. Conclusions: The Turkish version of the SCALE appears to be a valid and reliable tool to assess selective voluntary motor control of the lower limbs in children with spastic CP. IMPLICATIONS FOR REHABILITATION: The Turkish version of the Selective Control Assessment of the Lower Extremity is a valid and reliable assessment for children with spastic CP. The SCALE scores differed significantly between Gross Motor Function Classification System levels I versus II and levels II versus III as well as between types of spastic CP. The current study suggests that the SCALE is a quick and easy outcome measure to assess selective motor control in patients with spastic CP.

PMID: 34994671

7. National Institutes of Health Pathways to Prevention Workshop: Physical Activity and Health for Wheelchair Users Jerry H Gurwitz, Noelle E Carlozzi, Kirsten K Davison, Kelly R Evenson, Darrell J Gaskin, Boris Lushniak

Arch Rehabil Res Clin Transl. 2021 Oct 17;3(4):100163. doi: 10.1016/j.arrct.2021.100163. eCollection 2021 Dec.

Health benefits of physical activity are well recognized in the general population for reducing the risk of chronic health

conditions. Less is known about the effects of physical activity on people currently using or who may use wheeled mobility devices in the future, specifically individuals with multiple sclerosis, cerebral palsy, and spinal cord injury who are at increased likelihood for use of a wheeled mobility device. On December 1-3, 2020, the National Institutes of Health convened the Pathways to Prevention workshop: "Can Physical Activity Improve the Health of Wheelchair Users?" to consider the available scientific evidence on the clinical benefits and harms of physical activity for people currently using or who may use wheeled mobility devices in the future, with the aim of developing recommendations to fill gaps in the evidence base. A multidisciplinary team of content area experts developed the agenda and an evidence-based practice center prepared the evidence report. An independent panel, selected by the National Institutes of Health, attended the workshop; convened to develop recommendations on the basis of the systematic review, presentations, and public comments received during the workshop; and revised recommendations based on public comments received. This final report summarizes the panel's findings and identifies current gaps in knowledge. The panel made recommendations for new research efforts, including novel methods and new research infrastructure to improve the evidence base about the effects of physical activity on people currently using or who may use wheeled mobility devices in the future.

PMID: 34977545

8. Vision Abnormalities in Children and Young Adults With Cerebral Palsy; A Systematic Review Samira Heydarian, Marziye Moradi Abbasabadi, Mehdi Khabazkhoob, Hosein Hoseini-Yazdi, Masoud Gharib

Semin Ophthalmol. 2022 Jan 3;1-9. doi: 10.1080/08820538.2021.2021248. Online ahead of print.

Aim: The current study was designed to provide detailed information on the prevalence of ocular abnormalities in patients with cerebral palsy (CP). Methods: Four international online scientific databases, including Web of Science, PubMed, Scopus, and Google Scholar were systemically searched. First, the titles of the articles were evaluated, and if relevant, their abstracts and full texts were reviewed. The quality of the studies was assessed using the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) checklist. Results: A total of 147 articles were found in the initial search. After applying the exclusion criteria, 65 articles were chosen for further review, from which 17 articles, comprising a total of 1734 patients with CP ranging in age from birth to 22 years, passed the STROBE quality check and were included in this review. The prevalence of ocular abnormalities in the CP patients reported in the evaluated studies ranged between 34% to 100%, with refractive error, strabismus, and nystagmus exhibiting the greatest overall prevalence at 52%, 48%, and 11%, respectively in this population. Conclusion: Early ocular assessment of children with CP is essential for an accurate diagnosis, personalized rehabilitation and performing early interventions to improve their visual function.

PMID: 34978933

#### 9. Pain Phenotypes in Adults Living With Cerebral Palsy

Mark D Peterson, Heidi J Haapala, Anna Kratz

Neurol Clin Pract. 2021 Dec;11(6):e848-e855. doi: 10.1212/CPJ.000000000001113.

Background and objectives: To identify pain phenotypes among adults living with cerebral palsy (CP) and compare phenotypes of pain intensity, anxiety and depressive symptoms, and self-reported perceived stress. Methods: Seventy-one adults with CP presented to the University of Michigan (mean age =  $39.3 \pm 16.2$ ; 43 women, 28 men). The median of 6 on the American College of Rheumatology fibromyalgia survey was used to classify patients for nociplastic pain centralization. The painDETECT Score was used to classify patients for neuropathic pain. These measures were then used to cross-classify each patient into 1 of 4 possible pain categories: neuropathic, nociplastic, mixed neuropathic/noclipastic, or nociceptive pain (-neuropathic/-nociplastic pain). Results: Twenty-eight adults with CP (39.4%) were classified as nociceptive, 24 (33.8%) as nociplastic, 8 (11.3%) as neuropathic, and 11 (15.5%) as mixed neuropathic/nociplastic. Subgroups differed significantly on average scores on the Brief Pain Inventory pain intensity scale, the Perceived Stress Scale, and on the Patient-Reported Outcomes Measurement Information System measures of anxiety and depression; the nociceptive pain subgroup reported lower pain and emotional distress compared with the other groups. Discussion: Findings suggest that type of pain is variable among adults with CP and may arise through multiple mechanisms.

## 10. Supporting Ultra Poor People with Rehabilitation and Therapy among families of children with Cerebral Palsy in rural Bangladesh (SUPPORT CP): Protocol of a randomised controlled trial

Mahmudul Hassan Al Imam, Israt Jahan, Mohammad Muhit, Manik Chandra Das, Rosalie Power, Arifuzzaman Khan, Delwar Akbar, Nadia Badawi, Gulam Khandaker

Randomized Controlled Trial PLoS One. 2021 Dec 31;16(12):e0261148. doi: 10.1371/journal.pone.0261148. eCollection 2021.

Introduction: Poverty is a key contributor to delayed diagnosis and limited access to early intervention and rehabilitation for children with cerebral palsy (CP) in rural Bangladesh. 97% of families of children with CP live below the poverty line in Bangladesh. Therefore, in low-and middle-income countries (LMICs), efforts to improve outcomes for children with CP (including health-related quality of life, motor function, communication, and nutritional attainments) should also include measures to improve family economic and social capital. We propose a randomised controlled trial (RCT) to evaluate the effectiveness of an integrated microfinance/livelihood and community-based rehabilitation (IMCBR) program for ultra-poor families of children with CP in rural Bangladesh. Material and methods: This will be a cluster RCT comparing three arms: (a) integrated microfinance/livelihood and community-based rehabilitation (IMCBR); (b) community-based rehabilitation (CBR) alone; and (c) care-as-usual (i.e. no intervention). Seven clusters will be recruited within each arm. Each cluster will consist of 10 child-caregiver dyads totalling 21 clusters with 210 dyads. Parents recruited in the IMCBR arm will take part in a microfinance/livelihood program and Parent Training Module (PTM), their children with CP will take part in a Goal Directed Training (GDT) program. The programs will be facilitated by specially trained Community Rehabilitation Officers. The CBR arm includes the same PTM and GDT interventions excluding the microfinance/livelihood program. The care-as-usual arm will be provided with information about early intervention and rehabilitation. The assessors will be blinded to group allocation. The duration of the intervention will be 12 months; outcomes will be measured at baseline, 6 months, 12 months, and 18 months. Conclusion: This will be the first RCT of an integrated microfinance/livelihood and CBR program for children with CP in LMIC settings. Evidence from the study could transform approaches to improving wellbeing of children with CP and their ultra -poor families.

PMID: 34972100

# 11. Development of hip displacement in cerebral palsy: a longitudinal register study of 1,045 children Philippe Wagner, Gunnar Hägglund

Acta Orthop. 2022 Jan 3;93:124-131. doi: 10.2340/17453674.2021.851.

Background and purpose - Hip surveillance in children with cerebral palsy (CP) includes repeated radiographic hip examinations and measurements of the hip migration percentage (MP) to identify hips in need of surgery early, to prevent dislocation with the fewest number of radiographic examinations possible. We analyzed the early development of the MP in hips operated on to prevent hip dislocation and hips stabilized without surgery Patients and methods - From the Swedish Surveillance Programme for CP, 5,899 radiographic measurements from 1,045 children with a Gross Motor Function Classification System level III-V born in 1996-2011 were analyzed. For children operated on to prevent hip dislocation, measurements up to the most recent preoperative radiograph were included. The hip with highest MP was analyzed for each child. A mixed-effects model was used to estimate the development of the MP at each age for each child and the population mean. Results - In the 702 children who did not undergo preventive surgery, the mean MP increased with decreasing velocity up to age 6 years. Here it reached 24% (95% confidence interval [CI] 24-25), with a velocity of 0.3%/year (CI 0.0-0.5), remaining approximately stable up to age 12 years. In the 343 children who underwent preventive surgery (219 adductor-psoas lengthening, 124 varus derotation osteotomy of proximal femur), the mean MP increased with an increasing velocity from a mean of 30% (CI 27-32) 3 years before the operation. Interpretation - An increasing rate of hip displacement in hips with an MP > 24% indicates the need for preventive surgery. Hips stabilized without preventive surgery had a decreasing displacement rate and were usually stabilized with an MP < 30% at age 6 years.

PMID: 34984476

12. The impact of birthweight on the development of cerebral palsy: A population-based matched case-control study Katarina Esih, Tamara Trunk, Damjan Osredkar, Ivan Verdenik, David Neubauer, Anja Troha Gergeli, Miha Lučovnik

Early Hum Dev. 2021 Dec 27;165:105533. doi: 10.1016/j.earlhumdev.2021.105533. Online ahead of print.

Background: Cerebral palsy (CP) is a common cause of physical impairment in children, especially in newborns who are small for gestational age (SGA). Aims: The aim of our study was to investigate the association between birth weight and the risk of developing CP, controlling for gestational age and plurality. Study design: This retrospective, observational, case-control study was based on Slovenian Registry of Cerebral Palsy (SRCP) and Slovenian National Perinatal Information System (NPIS) data for the period 2002 to 2010. Subjects: For each pregnancy that resulted in the birth of the newborn(s) who later developed CP (n = 254), three pregnancies with newborns who did not develop CP (n = 762) were selected and matched for gestational age and plurality. Outcome measures: Diagnosis of CP was made at age 5 years or older by a developmental pediatrician trained in child neurology or a child neurologist using standard measures. Results: Risk of CP increased progressively as birth weight percentiles fell below the 50th centile, with children in the lowest percentiles at greatest risk. Birth weight percentiles traditionally classified as SGA were an independent risk factor for developing CP, with an odds ratio of 2.43 (95% confidence interval 1.57, 3.73). Conclusions: The results of this study suggest that the risk for developing CP is inversely related to birth weight, even at birth weights that do not meet the standard definitions of SGA. Synopsis - study question: Does birth weight represent a potential risk factor for the development of cerebral palsy (CP) when controlling for gestational age and plurality? What's already known: Newborns who are small for gestational age (SGA) are at higher risk of developing CP according to published studies. However, different definitions of SGA (birth weight below the 10th, 5th, or 3rd percentile for gestational age) have been used by researchers and clinicians, making it difficult to compare studies. What this study adds: This study suggests that the risk of developing CP is inversely related to birth weight, even at birth weights that do not meet standard definitions of SGA.

PMID: 34973634

## 13. Long-term outcomes of very-low-birth-weight and low-birth-weight preterm newborns with neonatal seizures: A single-center perspective

S C Schüssler, M Schmidt, L Deiters, A Candova, F B Fahlbusch, R Trollmann

Eur J Paediatr Neurol. 2021 Dec 22;36:137-142. doi: 10.1016/j.eipn.2021.12.013. Online ahead of print.

Objective: Newborn seizures are frequent in preterm newborns and indicate brain lesions in many cases. The objective of this observational study was to investigate the long-term outcome of very-low-birth-weight (VLBW) and low-birth-weight (LBW) preterm infants with neonatal seizures. Methods: We examined 54 preterm infants (40 VLBW and 14 LBW cases) born between 2008 and 2011 with clinical seizures during the neonatal period confirmed by interictal or ictal electroencephalography recordings in a retrospective single-center study. Neurodevelopmental follow-up included an expert neurological examination and cognitive testing (Kaufman Assessment Battery for Children) at a mean age of six years. Results: The (mean  $\pm$  standard deviation) gestational ages of the VLBW and LBW infants were  $27.2 \pm 1.9$  weeks and  $33.4 \pm 1.7$  weeks, respectively, and the postnatal age at seizure onset was  $13 \pm 11$  days in VLBW infants and  $9 \pm 8$  days in LBW infants, with a wide range of one to 62 days. LBW infants more frequently developed non-motor seizures (50.0%) than VLBW infants did (25.0%), and higher-grade intracranial hemorrhage was the predominant etiology in the VLBW group (18.0%), while the etiology in the LBW group was more heterogeneous and included central nervous system malformations and genetic syndromes. At the mean age of  $6.2 \pm 2.0$ , years, 25/54 patients were assessed and 44.4% of the VLBW group and 71.4% of the LBW group showed intellectual impairment. Infantile cerebral palsy was present in 22% of VLBW and 42.9% of LBW infants, respectively. Significance: The present analysis of long-term neurodevelopmental outcomes of preterm neonates who experienced seizures shows that the risk for intellectual impairment is not limited only to VLBW infants but may significantly affect LBW infants as well. The etiological spectrum differs in relation to gestational age.

PMID: 34973622

## 14. Systematic review and meta-analysis: the effect of bronchopulmonary dysplasia on neurodevelopment in very low birth weight premature infants

Shuqiang Gao, Xiaolong Zhang, Weina Du, Xiaofeng Zhou, Yufeng Xi, Rong Ju

Transl Pediatr. 2021 Nov;10(11):3023-3033. doi: 10.21037/tp-21-449.

Background: A meta-analysis was performed to study the effect of steroid intervention on the neurodevelopment of extremely low birth weight preterm infants complicated with bronchopulmonary dysplasia, and to provide a theoretical basis for clinical

treatment. Methods: The Wanfang database, Chinese Biomedical Literature database, VIP database, Baidu Academic, CNKI database, The Cochrane Library, Medline, Embase, and PubMed database were searched by computer from establishment to 2021. Randomized controlled trials on the effect of steroids on neurodevelopment in very low birth weight preterm infants with bronchial dysplasia published from January 10, 2007 were retrieved. The included literature was evaluated for bias risk, then analyzed using RevMan 5.3 software. Results: A total of 9 studies were included, with a total of 2,453 patients. The funnel plot showed that the circles and the midline of some studies were basically symmetrical, and there was no bias in the publications. The conclusions obtained were relatively reliable. Cerebral palsy, neurodevelopmental indicators, and MRI findings of preterm infants were analyzed. The cognitive impairment of very low birth weight preterm infants complicated with bronchial dysplasia (RR =0.83, 95% CI: 0.72-0.96, P=0.01) in the treatment group was significantly different from that in the control group, while cerebral palsy (RR =0.99, 95% CI: 0.75-1.29, P=0.93), speech impairment (RR =0.75, 95% CI: 0.46-1.21, P=0.24), hearing loss requiring amplification (RR =0.60, 95% CI: 0.35-1.03, P=0.06), bilateral blindness RR =0.81, 95% CI: 0.52-1.24, P=0.32), severe intraventricular hemorrhage (IVH) (RR =0.71, 95% CI: 0.33-1.50, P=0.37), and cystic periventricular leukomalacia (RR =0.82, 95% CI: 0.43-1.57, P=0.56) had no significant differences compared with the control group. Discussion: In this meta-analysis, we found that the use of steroids in very low birth weight preterm infants complicated with bronchial dysplasia had significant effects on cognition, but no significant effects on hearing, vision, or language function.

PMID: 34976768

**15. Neonatal brain injury influences structural connectivity and childhood functional outcomes**Alice Ramirez, Shabnam Peyvandi, Stephany Cox, Dawn Gano, Duan Xu, Olga Tymofiyeva, Patrick S McQuillen

PLoS One. 2022 Jan 5;17(1):e0262310. doi: 10.1371/journal.pone.0262310. eCollection 2022.

Neonatal brain injury may impact brain development and lead to lifelong functional impairments. Hypoxic-ischemic encephalopathy (HIE) and congenital heart disease (CHD) are two common causes of neonatal brain injury differing in timing and mechanism. Maturation of whole-brain neural networks can be quantified during development using diffusion magnetic resonance imaging (dMRI) in combination with graph theory metrics. DMRI of 35 subjects with CHD and 62 subjects with HIE were compared to understand differences in the effects of HIE and CHD on the development of network topological parameters and functional outcomes. CHD newborns had worse 12-18 month language (P<0.01) and 30 month cognitive (P<0.01), language (P = 0.05), motor outcomes (P = 0.01). Global efficiency, a metric of brain integration, was lower in CHD (P = 0.03) than in HIE, but transitivity, modularity and small-worldness were similar. After controlling for clinical factors known to affect neurodevelopmental outcomes, we observed that global efficiency was highly associated with 30 month motor outcomes (P = 0.02) in both groups. To explore neural correlates of adverse language outcomes in CHD, we used hypothesisbased and data-driven approaches to identify pathways with altered structural connectivity. We found that connectivity strength in the superior longitudinal fasciculus (SLF) tract 2 was inversely associated with expressive language. After false discovery rate correction, a whole connectome edge analysis identified 18 pathways that were hypoconnected in the CHD cohort as compared to HIE. In sum, our study shows that neonatal structural connectivity predicts early motor development after HIE or in subjects with CHD, and regional SLF connectivity is associated with language outcomes. Further research is needed to determine if and how brain networks change over time and whether those changes represent recovery or ongoing dysfunction. This knowledge will directly inform strategies to optimize neurologic functional outcomes after neonatal brain injury.

PMID: <u>34986206</u>

16. Evaluating the Effects of 5-Hz Repetitive Transcranial Magnetic Stimulation With and Without Wrist-Ankle Acupuncture on Improving Spasticity and Motor Function in Children With Cerebral Palsy: A Randomized Controlled Trial

Jiamin Li, Cen Chen, Shenyu Zhu, Xiulian Niu, Xidan Yu, Jie Ren, Min Shen

Front Neurosci. 2021 Dec 15;15:771064. doi: 10.3389/fnins.2021.771064. eCollection 2021.

Objective: The goal of this study is to explore the effect of wrist-ankle acupuncture combined with 5-Hz repetitive transcranial magnetic stimulation (rTMS) on improving spastic state and motor function of children with spastic cerebral palsy by measuring electrophysiological parameters and behaviors. Methods: Twenty-five children with spastic cerebral palsy were enrolled in a single-blind and randomized controlled trial. The control group received 20 sessions of 5-Hz rTMS over the affected hemisphere with 1,000 pulses. The experimental group was given wrist-ankle acupuncture on the basis of the control group. Gross motor function measure (GMFM-66), muscle tension, and electrophysiological parameters of the two groups were

assessed at baseline and after intervention. Results: After treatment, the GMFM-66 scores in the same groups were significantly improved (p < 0.001). Besides, the R-value of soleus, gastrocnemius, and hamstring muscle decreased (p < 0.05), and the results showed a trend of shortening MEP latency, increasing amplitude and duration (p < 0.05). Compared to the controlled group, the experimental group displayed more excellent changes in the GMFM-66 scores and motor evoked potential (MEP) latency. The statistical results showed that the increase of GMFM-66 score and the shortening of MEP latency in the experimental group were greater than that in the control group (p < 0.05). However, no significant differences were found in the assessment of muscle tension, amplitude, and duration of MEPs between two groups (p > 0.05). Conclusion: Wrist -ankle acupuncture combined with 5-Hz rTMS is optimal to improve gross motor function and enhance the conductivity of corticospinal tract in children with cerebral palsy but cannot highlight its clinical superiority in improving spasticity. Clinical Trial Registration: [http://www.chictr.org.cn/index.aspx], identifier [chictr2000039495].

PMID: 34975377

#### 17. Looking beyond motor function-adaptive behaviour in children with unilateral spastic cerebral palsy Sebastian Grunt

Editorial Eur J Paediatr Neurol. 2021 Dec 30;S1090-3798(21)00236-1. doi: 10.1016/j.ejpn.2021.12.016. Online ahead of print.

PMID: 34991946

#### 18. Social Outcomes of School Leavers With Cerebral Palsy Living in Victoria

Christine Imms, Dinah Reddihough, Daisy A Shepherd, Anne Kavanagh

Front Neurol. 2021 Dec 14;12:753921. doi: 10.3389/fneur.2021.753921. eCollection 2021.

Objective: In Australia, the National Disability Strategy provides a framework to guide actions and investment to achieve equity in social inclusion and economic participation for people with disability. We investigated the social outcomes of school leavers with cerebral palsy (CP) in Victoria, Australia and explored the determinants of desirable outcomes. Methods: We used the Victorian CP Register to invite all adults with CP aged 18-25 years (n = 649). On-line and/or paper-based surveys explored participation in education, employment, community activities, living situation, relationships and life satisfaction. Functional and health status data were collected. Social outcomes were summarized descriptively and compared between individuals with CP and non-disabled peers aged 18-25 years from the Household Income and Labor Dynamics in Australia dataset. Within the CP cohort we explored whether physical and mental health and level of functioning were associated with social outcomes. In addition, a descriptive comparison was undertaken between the social outcomes of the current CP cohort with that of a previously reported 2007 cohort. Results: Ninety participants (57% male; mean age 22.4 years (SD: 2.2) in 2020; 61.1% selfreported) provided data for analyses; response rate 16.9%. CP characteristics were similar between respondents and nonrespondents. In comparison to similar aged peers, 79.8% had completed secondary school (compared to 83.2%); 32.6% (compared to 75.8%) were in paid work; 87.5% (compared to 48.2%) were living in their parental home; and 3.4% (compared to 31.6%) were married or partnered. Individuals with CP and higher levels of functional capacity and better physical health were more likely to undertake post-secondary education. Higher levels of functional capacity and physical health, as well as lower mental health status were associated with being employed. Conclusions: While foundational education completion rates were similar to non-disabled peers, significant gaps in social outcomes remain, including residence in the parental home and single status. While addressing these issues is challenging, substantial efforts are needed to reduce these disparities-work that needs to be done in collaboration with people with CP and their families.

PMID: 34970206

#### 19. Tetrasomy 18p Initially Misdiagnosed as Cerebral Palsy in an Adult Patient

Yusuf Mehkri, Rebecca Jules, Aisha Elfasi, Hans Shuhaiber

Case Reports Cureus. 2021 Nov 30;13(11):e20053. doi: 10.7759/cureus.20053. eCollection 2021 Nov.

Tetrasomy 18p is a rare genetic condition characterized by a supernumerary 18p isochromosome with two copies of the p arm of chromosome 18 causing patients to have an extra chromosome. Most cases are de novo; however, a few maternally inherited

cases have been reported. The most commonly reported manifestations of this condition are developmental delay, cognitive impairments, muscle tone abnormalities, and dysmorphic facial features. This case details a new diagnosis of tetrasomy 18p in a 42-year-old adult who was initially diagnosed with cerebral palsy as a child. We compare the phenotypic traits of our patient with the ones reported in the literature.

PMID: 34993029

#### 20. Birth outcomes among women with congenital neuromuscular disabilities

Michelle Huezo García, Samantha E Parker, Julie M Petersen, Eric Rubenstein, Martha M Werler

Disabil Health J. 2021 Dec 1;101259. doi: 10.1016/j.dhjo.2021.101259. Online ahead of print.

Background: Women with disabilities are at an increased risk for adverse birth outcomes; however, research among women with congenital neuromuscular disabilities (CNMD) is limited. Objective: To describe characteristics and compare birth outcomes among mothers with and without cNMD. Methods: Data were from the Slone Birth Defects Study (case-control, conducted from 1976 to 2015), which collected information on demographic, reproductive, and lifestyle characteristics. cNMD included spina bifida, cerebral palsy, muscular dystrophy, contractures, or arthrogryposis and were identified by participant report. Those with cNMD were matched to participants without cNMD by interview year and study site. We use modified Poisson regression to estimate relative risks (RR) for low birthweight, macrosomia, preterm birth, and small/large-forgestational age (SGA/LGA) Given the case-control design and overrepresentation of infants with congenital anomalies, data were weighted to reflect a 3% national prevalence of infants with congenital anomalies. Results: Mothers with cNMD (n = 125) were more likely to be white, nulliparous, have a cesarean section, have an unplanned pregnancy, report a pre-pregnancy BMI ≥25 kg/m2, smoke during pregnancy, and report genitourinary infections. Mothers with cNMD had infants with shorter gestational length (mean difference: -7.44 days, 95% CI: -13.94, -0.95) compared to mothers without cNMD. cNMD was associated with higher risk of preterm birth (RR = 3.98, 95% CI: 1.33, 11.95) and SGA (RR = 2.14, 95% CI: 0.74, 6.15). Conclusion: Mothers with cNMD were more likely to deliver preterm and have an SGA infant. These findings highlight disparities faced by mothers with cNMD and stress the need to provide optimal perinatal and reproductive care.

PMID: 34980574

## 21. Understanding the Lived Experience of Caring for a Child with Severe Cerebral Palsy: A Critical Step toward Psychologically Informed Family-Centered Care

Jason E Cook, Melissa M Tovin, Melissa M Tovin

Phys Ther. 2021 Dec 23;pzab294. doi: 10.1093/ptj/pzab294. Online ahead of print.

Objective: The purpose of this study was to explore the lived experience of parents who care for children with cerebral palsy who function at a Gross Motor Function Classification System Level V (CP GMFCS Level V), their beliefs about pain and non -pain related emotional distress, and what it means to provide care and comfort. Methods: A phenomenological research design was used. Eleven participants were involved in this study. Pilot interviews informed the interview guide. Data were collected via participant journals and semi-structured interviews, and analyzed using van Manen's framework for understanding experience. Trustworthiness criteria were met through a variety of strategies to ensure a rigorous research process. Results: Four themes were revealed in this study: "Life is Hard, Heavy with Burden, Worry, and Love", "Remarkable", "Identity Transformation and Empowerment", and "Living a Life that is Planned, Forced with Structure and without Spontaneity." Conclusion: Caring for a child with lifelong needs is complex and requires a sensitive awareness of the contextual factors that impact daily decisions and routines. Understanding the lived experiences of parents who care for children with CP GMFCS Level V is necessary to provide psychologically informed, family-centered care. Comfort theory is presented as a framework for understanding what factors influence comfort and well-being. Understanding the complex nature of comfort for an individual can lead to greater understanding and empathy driven care. These results will provide a foundation for future studies that aim to enhance pediatric physical therapist care through provider empathy and understanding. Impact: Caregiving parent experiences are impactful and play a large role in the life of children with developmental disabilities. Research exploring the lived experience of caregivers may enhance empathy-driven, psychologically informed, family-centered physical therapist care throughout the life course. Lay summary: If you are a parent who cares for a child with severe cerebral palsy, you have unique experiences that impact day-to-day activities and lifelong planning for your family and child.

### 22. Perceived impact of lockdown on daily life in children with physical disabilities and their families during the COVID-19 pandemic

Roxane Varengue, Sylvain Brochard, Sandra Bouvier, Rodolphe Bailly, Laetitia Houx, Mathieu Lempereur, Christèle Kandalaft, Alain Chatelin, Jacky Vagnoni, Carole Vuillerot, Vincent Gautheron, Elea Dheilly, Christelle Pons, Mickael Dinomais, Marine Cacioppo

Child Care Health Dev. 2021 Dec 28. doi: 10.1111/cch.12952. Online ahead of print.

Background: The first lockdown during COVID-19 pandemic in France led to an abrupt change in children's daily lives. For children with physical disabilities and their families, activities were limited, access to healthcare and therapy was disrupted, and family organization was altered. The objective was to report the impact of the lockdown on daily life activities and wellbeing of children with physical disabilities as perceived by caregivers. Methods: Two online national surveys were addressed to the parents of children with physical disabilities (ECHO survey: 6 April to 11 May 2020) and without disabilities (E-COPAIN survey: 24 April to 11 May 2020), confined at home during the lockdown. A lockdown impact score was calculated from difficulties related to children's well-being (morale, behaviour and social interaction) and daily life activities (schooling and physical activity) and compared between groups. Data on family environment, parental stress and concerns were collected. Results: One thousand three hundred seventy-six children (9.45 ± 4.78 years, 54% girls) in ECHO survey and 367 children (7.3 ± 4.4 years, 48% girls) in E-COPAIN survey were included. A negative impact of lockdown was found on 81% of children with physical disabilities. Behavioural problems were significantly more frequent (59.5% vs. 47.4%, P < .005) and parental stress was higher  $(6.1 \pm 3.33 \text{ vs. } 5.3 \pm 3.01, P = .005)$  in the ECHO group. Associated impairments (odds ratio [OR] = 1.45 [1.30-1.62], P < .001), parental stress (OR = 1.09 [1.06-1.12], P < .001) and continuation of rehabilitation (OR = 0.80 [0.72-0.89], P < 0.001) were determinants of the level of difficulty experienced. Conclusions: The lockdown had a considerable, negative impact on the daily life of children with disabilities and their families. Guiding policymakers with the essential daily life activities and the services to provide for children with physical disabilities would offer valuable insights to manage such a sanitary crisis and allow to identify the most vulnerable population.

PMID: 34964148

#### 23. Anaesthesia and cerebral palsy

H Hayakawa, E S Pincott, U Ali

Review BJA Educ. 2022 Jan;22(1):26-32. doi: 10.1016/j.bjae.2021.08.003. Epub 2021 Nov 15.

PMID: 34992798

#### 24. Movement disorders, cerebral palsy and vaccination

Zuzana Liba, Josef Kraus, Tomas Necas, Jiri Necas, Miloslav Klugar, Pavel Krsek

Eur J Paediatr Neurol. 2021 Dec 21;36:143-150. doi: 10.1016/j.ejpn.2021.12.006. Online ahead of print.

This review focused on vaccination in children with movement disorders, including cerebral palsy and the movement disorders triggered by vaccination in children with and without neurological disabilities. The following clinical questions were addressed: 1) Can children with movement disorders be vaccinated? 2) Can vaccination trigger movement disorders in children without neurological disabilities? 3) Can vaccination trigger movement disorders in children with neurological disabilities? and 4) Is there any consensus of care concerning vaccination in children with movement disorders? Following the PRISMA reporting guidelines, 1096 records were identified and 34 relevant papers were included. No evidence that vaccinations are contraindicated for children with movement disorders was noticed. Several reports of neurological adverse events, including movement disorders in children without neurological disabilities after various types of vaccination, were found. The reporting rates were low, the causality was controversial, and patient outcomes were mostly favourable. There was limited (if any) evidence in our search that any vaccination leads to any movement disorder exacerbation. Finally, no generally accepted consensus or standards of care concerning vaccination in patients with movement disorders were found. In summary, we found few precautions for vaccination in this group of patients and concluded that general best practice guidelines for immunization should be followed. In addition, influenza and pneumococcal vaccines are recommended because they can reduce morbidity and mortality in individuals severely affected by movement restrictions.