

# Cerebral palsy research news

Monday 18 March 2024

**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 <a href="mailto:cerebralpalsy.org.au/our-research">cerebralpalsy.org.au/our-research</a>

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

Subscribe to CP Research News

### **Interventions and Management**

1. Critically appraised paper: Hand-arm bimanual intensive therapy including lower extremities (HABIT-ILE) improves bi-manual performance and gross motor function in pre-school children with unilateral cerebral palsy [synopsis]

Nikki Milne

J Physiother. 2024 Mar 11:S1836-9553(24)00004-3. doi: 10.1016/j.jphys.2024.02.005. Online ahead of print.

No abstract available

PMID: 38472054

2. Critically appraised paper: Hand-arm bimanual intensive therapy including lower extremities (HABIT-ILE) improves bimanual performance and gross motor function in pre-school children with unilateral cerebral palsy [commentary]

Sarah Reedman

J Physiother. 2024 Mar 11:S1836-9553(24)00005-5. doi: 10.1016/j.jphys.2024.02.004. Online ahead of print.

No abstract available

PMID: 38472051

3. Measuring Upper Extremity Activity of Children With Unilateral Cerebral Palsy Using Wrist-Worn Accelerometers: A Pilot Study

Sudha Srinivasan, Nidhi Amonkar, Patrick D Kumavor, Deborah Bubela

Observational Study Am J Occup Ther. 2024 Mar 1;78(2):7802180050. doi: 10.5014/ajot.2024.050443.

Importance: Children with unilateral cerebral palsy (UCP) have poor strength and movement control on one side of their body, leading to impaired bimanual coordination skills. Objective: To compare duration and intensity of all-day habitual movement of the dominant and nondominant upper extremities (UEs) in typically developing (TD) children and children with UCP. Design: Two-group observational study. Setting: Children's naturalistic settings. Participants: Convenience sample of 9 TD children and 9 children with UCP. Intervention: Children wore accelerometers on both wrists all day and night for 1 wk. Outcomes and measures: We compared the extent of asymmetry in bilateral arm use (intensity and duration) between the 2 groups. Results: Compared with TD children who use both UEs equally, children with UCP were more likely to use their dominant or unaffected UE than their nondominant or affected UE during daily activities. There were no differences between groups in dominant UE activity. However, children with UCP engaged in lower levels of moderate to vigorous activity and greater levels of light activity with their nondominant or affected UE than their TD peers. Conclusions and relevance: Wrist-

worn accelerometry can provide objective information on real-world habitual activity with both arms in children. Accelerometers are nonintrusive, easy to use, and well tolerated by children, and they allow prolonged monitoring of UE activity outside therapeutic contexts. Occupational therapists can use wrist-worn accelerometers as sensitive tools to assess asymmetries in UE use at baseline and as an outcome measure to assess the efficacy of behavioral interventions and carryover into real-world settings among children with UCP. Plain-Language Summary: This pilot study provides promising evidence that supports the use of wrist-worn accelerometry as an accurate, easy-to-use, and objective assessment tool for children with unilateral cerebral palsy (UCP) to detect asymmetries in bilateral real-world arm activity at baseline and after intensive occupational therapy interventions to improve arm function. The authors used wrist-worn accelerometry for one week with 9 typically developing (TD) children and 9 children with UCP to compare dominant or unaffected versus nondominant or affected upper extremity (UE) use for intensity and duration of activity. Compared with TD children, children with UCP had lower relative intensity of activity in the nondominant UE than the dominant UE. Wrist-worn accelerometers seem to be a sensitive measure to detect asymmetries in bilateral all-day UE use in children with UCP. The findings have implications for the use of wrist-worn accelerometers as an outcome measure to assess the efficacy of intensive therapies to improve real-world affected UE activity and bimanual function among children with UCP.

PMID: 38478583

## 4. Outcome of Open Hip Reduction, Pelvic Osteotomy, and Varus Derotational Osteotomy in Children With Cerebral Palsy: A Retrospective Study

Ozair Bin Majid, Zayed S Alzayed, Iram Saba, Alia A Aournaser, Ruby Anne A Valoria, Saeed Koaban, Shahad A Zaabi, Alaeldein A Nogud, Abdulrahman M Sharif

Cureus. 2024 Feb 11;16(2):e53996. doi: 10.7759/cureus.53996. eCollection 2024 Feb.

Introduction For spastic hip dislocations, a variety of operations are available with open hip reduction and varus derotational osteotomy of the proximal femur combined with pelvic osteotomy ± adductor release being a good option with favourable outcomes. This study aims to assess the outcome and complications of combined open hip reduction with pelvic osteotomy and varus derotational osteotomy. Methods In this study, 70 hips in 52 patients with spastic hip dislocation due to cerebral palsy were included. All included patients were treated surgically in our institute between January 2016 and December 2021. There were 31 males and 21 females. For each patient, information was collected about the age at the time of surgery and different radiological parameters at three different time intervals: pre-operatively, immediately post-operatively, and at the final followup. We also collected information about any complications arising from the surgery performed. Results The mean duration of follow-up was 19.58 months. The acetabular index decreased from an average of 35.01° to 17.18° with a mean difference of 17.83° (p<0.001). The central edge angle, which averaged -49.13° in the pre-operative period, increased to 26.34° and then marginally decreased to 25.47° at the final follow-up. The average migration index of 80.51% in the pre-operative period improved to 1.4% post-operatively with a mean difference of -79.11% (p<0.01). The migration index increased to 8.54% at the final follow-up. Similarly, the neck-shaft angle, which averaged 160.89° in the pre-operative period, decreased to 125.23° at the time of final follow-up with a percentage change of -22.16%. Conclusion Single-stage combined surgery in the form of combined open hip reduction and pelvic osteotomy with varus derotational osteotomy successfully treats the condition and shows good outcomes in patients with spastic hip dislocations. This treatment is associated with very few complications.

PMID: 38476797

## 5. Treatment of Hip Displacement in Children With Cerebral Palsy: A 5-year Comparison of Proximal Femoral Osteotomy and Combined Femoral-Pelvic Osteotomy in 163 Children

Nikolaos Kiapekos, Johan von Heideken, Eva Broström, Gunnar Hägglund, Per Åstrand

J Pediatr Orthop. 2024 Mar 13. doi: 10.1097/BPO.000000000002674. Online ahead of print.

Background: This study compared the 5-year outcomes of isolated proximal femoral varus osteotomy (FO) and combined proximal femoral varus and pelvic osteotomy (FPO) for the treatment of hip displacement in children with cerebral palsy (CP) in Sweden, focusing on the number of reoperations and residual hip displacement. Methods: The study included 163 children with a 5-year follow-up after FO or FPO in the national Swedish CP surveillance program, CPUP. Descriptive statistics and univariate and multivariate Cox regression analyses were used to identify whether the age at surgery, sex, Gross Motor Function Classification System level, CP dominant symptom, hip migration percentage (MP), type of surgery (unilateral/bilateral), and history of soft tissue hip surgery were related to the 5-year outcomes after surgery. Failure after hip surgery was defined as a skeletal reoperation involving the hip and/or MP >50%. Results: During the period 2001 to 2017, 163 children (65 girls) underwent 246 femoral and/or pelvic osteotomies (154 FO, 47 bilaterally; 92 FPO, 16 bilaterally) and had a 5-year follow-up; 95 and 74 children had ≥1 FO or FPO as the primary skeletal surgery, respectively. The mean preoperative MP (51%±18% for FO and 59%±17% for FPO, P=0.001) and age at surgery (6.2±2.5 years for FO and 7.3±2.8 years for FPO, P=0.014) differed between procedures. At the 5-year follow-up, 5 hips (5%) had reoperations and 5 hips (5%) had radiological failure among the 92 FPOs, and 33 (21%) had reoperations and 14 (9%) radiological failure among the 154 FOs. The difference in outcome failure rate was significant (P<0.001). Multivariate Cox regression analysis showed a lower risk for failure with FPO

[hazard ratio (HR)=0.32, 95% CI: 0.15-0.68] compared with FO. A higher preoperative MP increased the risk for outcome failure (HR=1.21, 95% CI: 1.15-1.36 for each 5% increment). Conclusions: FPO had a higher mean preoperative MP but a lower 5-year outcome failure rate compared with FO. A higher preoperative MP was associated with an increased risk of failure. Level of evidence: Level II-prospective comparative study.

PMID: 38477355

## 6. Quality of life and clinical outcomes in severely involved cerebral palsy patients and spastic hips undergoing Castle surgery: a cross-sectional study

Helder Henzo Yamada, Patricia Maria de Moraes Barros Fucs

Int Orthop. 2024 Mar 14. doi: 10.1007/s00264-024-06135-2. Online ahead of print.

Purpose: As progressive hip dislocation causes pain in children with spastic cerebral palsy (CP) and spasticity needs surgical correction, we aimed to describe clinical and radiographic outcomes in CP patients with painful hip deformity treated with the Castle salvage procedure. Methods: We included all patients operated in the same hospital between 1989 and 2017 with painful spastic hips and femoral head deformity making joint reconstruction unfeasible. We collected clinical and functional data from medical records and evaluated radiographies to classify cases for femoral head shape and migration, type of deformity, spinal deformity, and heterotopic ossification. We investigated quality of life one year after surgery. Results: We analyzed 41 patients (70 hips) with complete medical records. All had severe function compromise GMFCS V (Gross Motor Function Classification System) and heterotopic ossifications, all but one had scoliosis, and most had undergone other surgeries before Castle procedure. Patients were followed up for 77.1 months (mean) after surgery. The mean initial migration index was 73%. Seven patients had complications, being three patients minor (two femur and one tibial fracture) and four majors (patients requiring surgical revision). Quality of life was considered improved by most of the carers (35 children; 85.3%) as level 4/5 according to CPCHILD instrument. No child was able to stand or walk, but moving in and out of bed, of vehicles, and to a chair, remaining seated, or visiting public places was "very easy." Conclusion: We considered most patients (37 patients-90%, 66 hips-94%) as having satisfactory outcomes because they had no or minor complications, absence of pain, free mobility of the lower limbs and were able to sit in a wheelchair.

PMID: 38483563

## 7. Gastrocnemius medialis and Achilles tendon properties do not differ between children with unilateral or bilateral spastic cerebral palsy

Andreas Habersack, Martin Svehlik, Bernhard Guggenberger, Markus Tilp, Annika Kruse

J Biomech. 2024 Mar 7:166:112041. doi: 10.1016/j.jbiomech.2024.112041. Online ahead of print.

Spastic cerebral palsy (SCP) is a common neurodevelopmental disorder in children, which can be categorized into unilateral and bilateral subtypes. Most studies examining the muscle-tendon properties of the lower extremities in individuals with SCP do not distinguish between subtypes. However, spastic muscle morphology is an important determinant for its function. Therefore, differences in muscle-tendon pathology might lead to different treatment strategies. The aim of this retrospective study was to investigate the muscle-tendon properties between children with unilateral SCP and those with bilateral SCP. Overall, 33 ambulatory children (15 with unilateral SCP and 18 with bilateral SCP, Gross Motor Function Classification System Level I-III) were included. Ankle joint range of motion, isometric muscle strength, and muscle-tendon properties of the gastrocnemius medialis (GM) muscle-tendon unit (MTU) (e.g., muscle volume, tissue lengthening behavior) were assessed with isokinetic dynamometry, 3D motion capture, and ultrasound, respectively. Independent t-tests or Mann-Whitney tests were used to test for group differences ( $\alpha = 0.05$ ). Effect sizes (Cohen's d) were also calculated. No significant differences in any assessed parameter were found between children with unilateral SCP and children with bilateral SCP (p > 0.05, d < 0.57). Our findings suggest that the functional and morphological properties of the GM MTU are similarly developed in children with unilateral SCP and children with bilateral SCP. We assume that activity levels might be the decisive factor. Nonetheless, our investigations need be extended by including gait parameters and associated tissue dynamics.

PMID: 38461743

## 8. Locomotion Efficiency in Children With Cerebral Palsy Experiencing Limited Gross Motor Function: Walking Versus Cycling

Cloé Dussault-Picard, Annie Pouliot-Laforte, Claire Cherriere, Eloïse Houle, Laurent Ballaz

Pediatr Phys Ther. 2024 Mar 1. doi: 10.1097/PEP.00000000001096. Online ahead of print.

Purpose: This study compares cycling and walking efficiency, and energy expenditure in children with bilateral spastic cerebral

palsy (CP). In children with CP, locomotion with body weight support aids such as a tricycle is a potential alternative for less exhausting movements. Methods: Nine children with CP traveled at comfortable speed for 6 minutes by cycling and walking. The energy expenditure index (EEI) and the percentage of the reserve heart rate (%HRR) were calculated. Results: The EEI was lower while cycling than walking, the traveled distance was higher while cycling than walking, and %HRR remained similar between cycling and walking. Conclusion: Cycling appears an efficient alternative to walking for children with CP for adapted school environments and in the community.

PMID: 38460146

#### 9. Under-recognition of leg dystonia in people with cerebral palsy

Bhooma Aravamuthan, Toni S Pearson, Keerthana Chintalapati, Keisuke Ueda

Ann Child Neurol Soc. 2023 Jun;1(2):162-167. doi: 10.1002/cns3.20018. Epub 2023 Apr 5.

Objective: To determine the rates of clinical under-documentation of leg dystonia in people with cerebral palsy (CP). Methods: In this prospective cohort study, we identified independently ambulatory people age 10-20yo with CP-associated spasticity seen in a tertiary care CP center between 1/1/20 to 11/4/21. Three pediatric movement disorders specialists assessed gait videos from these visits for leg dystonia using the Global Dystonia Rating Scale. We compared the gold standard expert consensus assessment for each patient with the clinical documentation of dystonia during a contemporaneous CP Center clinic visit and also with dystonia documentation longitudinally in their medical record. Results: Of 116 people with CP-associated spasticity assessed in this study, 70 were found to have leg dystonia in their gait videos. Only 13% of these 70 individuals (n=9/70) had leg dystonia documented in their contemporaneous CP Center clinic visit, even though they were assessed during this visit by clinicians well-trained in CP and dystonia assessment. Even with repeated assessment, only 54% (n=38/70) of these individuals had leg dystonia documented in their medical record. Conclusions: Leg dystonia is clinically under-documented in people with CP-associated spasticity, even when these people are evaluated by well-trained clinicians. Longitudinal evaluation and vigilance for leg dystonia is critical to address this diagnostic gap.

PMID: 38464792

## 10. The effects of neurofeedback training for children with cerebral palsy and co-occurring attention deficits: A pilot study

Yuh-Chuan Chen, Wen-Pin Chang, Kai-Jie Liang, Chia-Ling Chen, Hsin-Yung Chen, Shu-Ping Chen, Pei-Ying S Chan

Randomized Controlled Trial Child Care Health Dev. 2024 Mar;50(2):e13231. doi: 10.1111/cch.13231.

Background: Limited research exists regarding the effectiveness of electroencephalogram (EEG) neurofeedback training for children with cerebral palsy (CP) and co-occurring attention deficits (ADs), despite the increasing prevalence of these dual conditions. This study aimed to fill this gap by examining the impact of neurofeedback training on the attention levels of children with CP and AD. Methods: Nineteen children with both CP and co-occurring ADs were randomly assigned to either a neurofeedback or control group. The neurofeedback group received 20 sessions of training, lasting approximately 1 h per day, twice a week. Theta/beta ratios of the quantitative electroencephalography (QEEG) recordings were measured pre-training and post-training in the resting state. The Continuous Performance Test (CPT), the Test of Visual Perceptual Skills-3rd Version (TVPS-3) and the Conners' Parent Rating Scale (CPRS) were measured at pre- and post-training. Results: The neurofeedback group showed both decreased theta/beta ratios compared with control group (p = 0.04) at post-training and a within-group improvement during training (p = 0.02). Additionally, the neurofeedback group had a trend of decreased omission rates of the CPT (p = 0.08) and the visual sequential memory and the visual closure subscores in the TVPS-3, compared with the control group (p = 0.02 and p = 0.01, respectively). Conclusions: The results suggested that children with CP and co-occurring AD may benefit from neurofeedback training in their attention level. Further research is needed to explore long-term effects and expand its application in this population.

PMID: 38465844

#### 11. Physical Activity in Cerebral Palsy: A Current State Study

Alberto J Molina-Cantero, Thais Pousada García, Soraya Pacheco-da-Costa, Clara Lebrato-Vázquez, Alejandro Mendoza-Sagrera, Paolo Meriggi, Isabel M Gómez-González

Healthcare (Basel). 2024 Feb 23;12(5):535. doi: 10.3390/healthcare12050535.

This document analyzes a survey conducted in three geographical areas in Spain, focusing on centers for individuals with cerebral palsy (CP). The study aims to determine the adherence rate to recommended physical activity guidelines, assess if there is a decline in interest in physical activity over time, identify the stage at which this decline occurs, and explore potential

mechanisms, tools, or strategies to sustain long-term engagement in regular physical activity for this population. The 36-item questionnaire comprises multiple-choice, open-ended, and Likert scale-type questions. Data were collected on physical activity frequency and duration, daily living activities, and demographics. Statistical analysis identified patterns and relationships between variables. Findings reveal that only a 17.6% meets the World Health Organization (WHO) recommendations regarding regular physical activity (RPA), decreasing in frequency or number of days a week, (3.7 d/w to 2.9 d/w; p < 0.01) and duration (50.5 min/d to 45.2 min/d; p < 0.001) with age, especially for those with higher Gross Motor Function Classification System (GMFCS) mobility levels. Obesity slightly correlates with session duration ( $\rho$  = -0.207; p < 0.05), not mobility limitations. Gender has no significant impact on mobility, communication, or physical activity, while age affects variables such as body mass index (BMI) and engagement (p < 0.01). A substantial proportion follows regular physical activities based on health professionals' advice, with interest decreasing with age. To improve adherence, focusing on sports-oriented goals, group sessions, and games is recommended. These findings emphasize the importance of personalized programs, particularly for older individuals and those with greater mobility limitations.

PMID: 38470646

## 12. Serious Game with Electromyography Feedback and Physical Therapy in Young Children with Unilateral Spastic Cerebral Palsy and Equinus Gait: A Prospective Open-Label Study

Christophe Boulay, Jean-Michel Gracies, Lauren Garcia, Guillaume Authier, Alexis Ulian, Maud Pradines, Taian Martins Vieira, Talita Pinto, Marco Gazzoni, Béatrice Desnous, Bernard Parratte, Sébastien Pesenti

Sensors (Basel). 2024 Feb 26;24(5):1513. doi: 10.3390/s24051513.

The clinical effects of a serious game with electromyography feedback (EMGs\_SG) and physical therapy (PT) was investigated prospectively in children with unilateral spastic cerebral palsy (USCP). An additional aim was to better understand the influence of muscle shortening on function. Thirty children with USCP (age  $7.6 \pm 2.1$  years) received four weeks of EMGs\_SG sessions  $2^{\times}$ /week including repetitive, active alternating training of dorsi- and plantar flexors in a seated position. In addition, each child received usual PT treatment  $\leq 2^{\times}$ /week, involving plantar flexor stretching and command strengthening on dorsi- and plantar flexors. Five-Step Assessment parameters, including preferred gait velocity (normalized by height); plantar flexor extensibility (XV1); angle of catch (XV3); maximal active ankle dorsiflexion (XA); and derived coefficients of shortening, spasticity, and weakness for both soleus and gastrosoleus complex (GSC) were compared pre and post treatment (t-tests). Correlations were explored between the various coefficients and gait velocities at baseline. After four weeks of EMGs\_SG + PT, there was an increase in normalized gait velocity from  $0.72 \pm 0.13$  to  $0.77 \pm 0.13$  m/s (p = 0.025, d = 0.43), a decrease in coefficients of shortening (soleus,  $0.10 \pm 0.07$  pre vs.  $0.07 \pm 0.08$  post, p = 0.004, d = 0.57; GSC  $0.16 \pm 0.08$  vs.  $0.13 \pm 0.08$ , p = 0.003, d = 0.58), spasticity (soleus  $0.14 \pm 0.06$  vs.  $0.12 \pm 0.07$ , p = 0.02, d = 0.46), and weakness (soleus  $0.14 \pm 0.07$  vs.  $0.11 \pm 0.07$ , p = 0.005, d = 0.55). At baseline, normalized gait velocity correlated with the coefficient of GSC shortening (R = -0.43, p = 0.02). Four weeks of EMGs\_SG and PT were associated with improved gait velocity and decreased plantar flexor shortening. A randomized controlled trial comparing EMGs\_SG and conventional PT is needed.

PMID: 38475049

## 13. Assessing the Match Physical Responses of International Referees for Footballers with Cerebral Palsy: A Tournaments and Halves Comparative Analysis

Matías Henríquez, Eñaut Ozaeta, Daniel Castillo, Raúl Reina, María Isabel Cornejo, Aitor Iturricastillo, Skye Arthur-Banning, Javier Yanci

Sensors (Basel). 2024 Feb 29;24(5):1595. doi: 10.3390/s24051595.

Similar to conventional football, the modality dedicated to footballers with cerebral palsy (CP) requires referees who cope with the physical demands imposed during competitive matches to apply the rules of the game. While a significant body of research has explored the physical demands on referees in mainstream football, there is a noticeable lack of data regarding CP football. This study aimed to examine the physical response of international referees participating in different levels of world competitions for footballers with CP. Thirteen international referees, who officiated 49 matches in the men's 2022 World Cup (1st to 15th ranked teams) and 2022 World Championships (16th to 30th ranked teams), participated in this study. A cross-sectional design was used to determine the physical responses and compare the 1st and 2nd halves and the performance in the different tournaments, recording physical variables throughout the matches. Significant higher physical responses were observed in the World Cup in comparison to the World Championship. Overall, high-level tournaments have been shown to elicit more intense physical responses from referees officiating CP football matches compared to lower-level tournaments. For the World Cup, a significantly higher number of accelerations and decelerations were registered in the 1st half compared to the 2nd half. This information may be useful for the strength and conditioning coaches of referees to plan weekly training sessions more specifically and adjust the periodical training load and post-match recovery protocols.

PMID: 38475129

#### 14. Cross-frequency cortex-muscle interactions are abnormal in young people with dystonia

Zhenghao Guo, Jean-Pierre Lin, Osvaldo Simeone, Kerry R Mills, Zoran Cvetkovic, Verity M McClelland

Brain Commun. 2024 Feb 26;6(2):fcae061. doi: 10.1093/braincomms/fcae061. eCollection 2024.

Sensory processing and sensorimotor integration are abnormal in dystonia, including impaired modulation of betacorticomuscular coherence. However, cortex-muscle interactions in either direction are rarely described, with reports limited predominantly to investigation of linear coupling, using corticomuscular coherence or Granger causality. Information-theoretic tools such as transfer entropy detect both linear and non-linear interactions between processes. This observational case-control study applies transfer entropy to determine intra- and cross-frequency cortex-muscle coupling in young people with dystonia/ dystonic cerebral palsy. Fifteen children with dystonic dystonic cerebral palsy and 13 controls, aged 12-18 years, performed a grasp task with their dominant hand. Mechanical perturbations were provided by an electromechanical tapper. Bipolar scalp EEG over contralateral sensorimotor cortex and surface EMG over first dorsal interosseous were recorded. Multi-scale wavelet transfer entropy was applied to decompose signals into functional frequency bands of oscillatory activity and to quantify intraand cross-frequency coupling between brain and muscle. Statistical significance against the null hypothesis of zero transfer entropy was established, setting individual 95% confidence thresholds. The proportion of individuals in each group showing significant transfer entropy for each frequency combination/direction was compared using Fisher's exact test, correcting for multiple comparisons. Intra-frequency transfer entropy was detected in all participants bidirectionally in the beta (16-32 Hz) range and in most participants from EEG to EMG in the alpha (8-16 Hz) range. Cross-frequency transfer entropy across multiple frequency bands was largely similar between groups, but a specific coupling from low-frequency EMG to beta EEG was significantly reduced in dystonia [P = 0.0061 (corrected)]. The demonstration of bidirectional cortex-muscle communication in dystonia emphasizes the value of transfer entropy for exploring neural communications in neurological disorders. The novel finding of diminished coupling from low-frequency EMG to beta EEG in dystonia suggests impaired cortical feedback of proprioceptive information with a specific frequency signature that could be relevant to the origin of the excessive low-frequency drive to muscle.

PMID: 38487552

## 15. Cerebral palsy and sleep: nonpharmacological treatment and impact on the life of caregivers - an integrative review [Abstract in English, Portuguese]

Marcela Fischer de Almeida, Suzane Mello, Marise Bueno Zonta, Ana Chrystina Crippa

Review Arq Neuropsiquiatr. 2024 Mar;82(3):1-9. doi: 10.1055/s-0044-1781464. Epub 2024 Mar 11.

Background: Children with cerebral palsy have a higher prevalence of sleep disorders, with numerous factors associated with a negative impact on the quality of life of caregivers. Objective: To identify factors related to sleep disorders, nonpharmacological treatment, and the impact on the lives of caregivers. Methods: The present literature review was carried out in the Latin American and Caribbean Center on Health Sciences Information (BIREME), the Cochrane Library, Scopus, PubMed, the Cumulative Index to Nursing and Allied Health Literature (CINAHL), PsycInfo, WorldCat, Web of Science, Latin American Literature on Health Sciences (LILACS), and Excerpta Medica Database (EMBASE), with the descriptors sleep, child, cerebral palsy, parents, and nursing. Studies available in Portuguese, English, or Spanish, published between 2010 and 2020, were our inclusion criteria. A total of 29 articles were included in the present review. Results: We considered nonpharmacological interventions effective support measures to drug-based treatments. The main sleep disorders in children with cerebral palsy are insomnia, parasomnias, nightmares, sleep bruxism, sleepwalking, sleep talking, disorders of initiation and maintenance of sleep, and sleep hyperhidrosis. Most studies point to a reduction in the quality of life of caregivers whose children have sleep disorders. Conclusion: Our review suggests the effectiveness of nonpharmacological treatments combined with the use of medications. Measures such as changes in sleep environment and routine are favorable strategies to improve sleep quality. In addition, children with sleep disorders negatively impact the quality of life of their caregivers.

PMID: 38467393

#### 16. Postoperative pain and pain management following selective dorsal rhizotomy

Isabel G Adams, Ramanie Jayaweera, Jennifer Lewis, Nadia Badawi, Mohamed E Abdel-Latif, Simon Paget

BMJ Paediatr Open. 2024 Mar 15;8(1):e002381. doi: 10.1136/bmjpo-2023-002381.

Background: Selective dorsal rhizotomy (SDR) is a neurosurgical procedure that reduces lower limb spasticity, performed in some children with spastic diplegic cerebral palsy. Effective pain management after SDR is essential for early rehabilitation. This study aimed to describe the anaesthetic and early pain management, pain and adverse events in children following SDR. Methods: This was a retrospective cohort study. Participants were all children who underwent SDR at a single Australian tertiary hospital between 2010 and 2020. Electronic medical records of all children identified were reviewed. Data collected included demographic and clinical data (pain scores, key clinical outcomes, adverse events and side effects) and medications

used during anaesthesia and postoperative recovery. Results: 22 children (n=8, 36% female) had SDR. The mean (SD) age at surgery was 6 years and 6 months (1 year and 4 months). Common intraoperative medications used were remifentanil (100%), ketamine (95%), paracetamol (91%) and sevoflurane (86%). Postoperatively, all children were prescribed opioid nurse-controlled analgesia (morphine, 36%; fentanyl, 36%; and oxycodone, 18%) and concomitant ketamine infusion. Opioid doses were maximal on the day after surgery. The mean (SD) daily average pain score (Wong-Baker FACES scale) on the day after surgery was 1.4 (0.9), decreasing to 1.0 (0.5) on postoperative day 6 (POD6). Children first attended the physiotherapy gym on median day 7 (POD8, range 7-8). Most children experienced mild side effects or adverse events that were managed conservatively. Common side effects included constipation (n=19), nausea and vomiting (n=18), and pruritus (n=14). No patient required return to theatre, ICU admission or prolonged inpatient stay. Conclusions: Most children achieve good pain management following SDR with opioid and ketamine infusions. Adverse events, while common, are typically mild and managed with medication or therapy. This information can be used as a baseline to improve postoperative care and to support families' understanding of SDR before surgery.

PMID: 38490692

#### 17. Non-immersive virtual reality based treatment for children with unilateral cerebral palsy: Preliminary results

Paolo Meriggi, Martina Mandalà, Mattia Randazzo, Elena Brazzoli, Anna Castagna, Valentina Di Giusto, Anna Cavallini, Alberto Marzegan, Tiziana Lencioni, Ivana Olivieri

J Pediatr Rehabil Med. 2024 Mar 15. doi: 10.3233/PRM-230028. Online ahead of print.

Purpose: Unilateral cerebral palsy (UCP) represents about 30-40% of overall cerebral palsy diagnoses. Upper limb impairment has a significant negative impact on activities of daily living (ADL), and recent studies have shown that the use of virtual reality (VR) can increase motivation and promote an improvement in ADL. This preliminary study was aimed at exploring the acceptability and usability of a VR rehabilitation treatment, using the VITAMIN Platform, for children with UCP. A secondary goal of the study was to compare the results of usual standardized clinical scales and questionnaires with kinematic results as well as with the quantitative measures acquired by the VITAMIN platform in each exercise of the rehabilitation sessions. Methods: Six children with UCP (aged 7-15) were recruited for a preliminary investigation in using a non-immersive VR system. The treatment was composed of 10 weekly sessions of 45 minutes. Each child played five types of exergames, using the impaired upper limb to hit virtual objects projected on a wide screen. Standardized clinical scales, kinematic analysis, and questionnaires were used to extensively assess upper limb function before and at the end of treatment. Five typicallydeveloping children provided a reference for the instrumented kinematic assessment. Results: At the end of the treatment, Melbourne Assessment 2 (MA2) scores increased for all the participants (mean increase in range of movement (ROM) + 19.1%, accuracy + 4.6%, dexterity + 13.1%, fluency + 10.3%). Shoulder flexion-extension ROM also improved (mean increase + 210.5°), and according to the kinematic analysis, shoulder movements became more similar to reference profiles. These results were confirmed by a general improvement in performing ADL, assessed by the ABILHAND-Kids questionnaire. Finally, a general agreement among the different measures and indexes emerged from the acquired data. Conclusion: The results show that VR treatment with the VITAMIN platform could be engaging and functional for rehabilitation of children with UCP. The good agreement among the qualitative and quantitative measures and indexes confirms the potential of such novel treatment. However, due to the limited sample size and small number of sessions, further and larger investigations are required to evaluate the effectiveness and to generalize the results.

PMID: 38489200

## 18. Prevalence, birth, and clinical characteristics of dyskinetic cerebral palsy compared with spastic cerebral palsy subtypes: A Norwegian register-based study

No authors listed

Dev Med Child Neurol. 2024 Mar 15. doi: 10.1111/dmcn.15905. Online ahead of print.

No abstract available

PMID: 38489770

## 19. Use of preventive service and potentially preventable hospitalization among American adults with disability: Longitudinal analysis of Traditional Medicare and commercial insurance

Elham Mahmoudi, Paul Lin, Dana Rubenstein, Timothy Guetterman, Amanda Leggett, Katherine L Possin, Neil Kamdar

Prev Med Rep. 2024 Feb 23:40:102663. doi: 10.1016/j.pmedr.2024.102663. eCollection 2024 Apr.

Objective: Examine the association between traditional Medicare (TM) vs. commercial insurance and the use of preventive care and potentially preventable hospitalization (PPH) among adults (18+) with disability [cerebral palsy/spina bifida (CP/SB);

multiple sclerosis (MS); traumatic spinal cord injury (TSCI)] in the United States. Methods: Using 2008-2016 Medicare and commercial claims data, we compared adults with the same disability enrolled in TM vs. commercial insurance [Medicare: n = 21,599 (CP/SB); n = 7,605 (MS); n = 4,802 (TSCI); commercial: n = 11,306 (CP/SB); n = 6,254 (MS); n = 5,265 (TSCI)]. We applied generalized estimating equations to address repeated measures, comparing cases with controls. All models were adjusted for age, sex, race/ethnicity, and comorbid conditions. Results: Compared with commercial insurance, enrolling in TM reduced the odds of using preventive services. For example, adjusted odds ratios (OR) of annual wellness visits in TM vs. commercial insurance were 0.31 (95% confidence interval (CI): 0.28-0.34), 0.32 (95% CI: 0.28-0.37), and 0.19 (95% CI: 0.17-0.22) among adults with CP/SB, TSCI, and MS, respectively. Furthermore, PPH risks were higher in TM vs. commercial insurance. ORs of PPH in TM vs. commercial insurance were 1.50 (95% CI: 1.18-1.89), 1.83 (95% CI: 1.40-2.41), and 2.32 (95% CI: 1.66-3.22) among adults with CP/SB, TSCI, and MS, respectively. Moreover, dual-eligible adults had higher odds of PPH compared with non-dual-eligible adults [CP/SB: OR = 1.47 (95% CI: 1.25-1.72); TSCI: OR = 1.61 (95% CI: 1.35-1.92), and MS: OR = 1.80 (95% CI: 1.55-2.10)]. Conclusions: TM, relative to commercial insurance, was associated with lower receipt of preventive care and higher PPH risk among adults with disability.

PMID: 38464419

## 20. Neurodevelopmental outcomes of extremely preterm infants with bronchopulmonary dysplasia (BPD) - A retrospective cohort study

Khoa L Nguyen, Dominic A Fitzgerald, Annabel Webb, Barbara Bajuk, Himanshu Popat

Review Paediatr Respir Rev. 2024 Mar 1:S1526-0542(24)00021-6. doi: 10.1016/j.prrv.2024.02.004. Online ahead of print.

Objective: To investigate the neurodevelopmental outcomes for preterm infants born < 29 weeks gestation with/without bronchopulmonary dysplasia (BPD). Study design: Preterm infants < 29 weeks' gestation born 2007-2018 in New South Wales and the Australian Capital Territory, Australia, were included. Infants who died < 36 weeks' postmenstrual age and those with major congenital anomalies were excluded. Subjects were assessed at 18-42 months corrected age using the Bayley Scales of Infant Development, 3rd edition. Results: 1436 infants without BPD (non-BPD) and 1189 infants with BPD were followed. The BPD group, 69 % infants were discharged without respiratory support (BPD1), 29 % on oxygen (BPD2) and 2 % on pressure support/tracheostomy (BPD3). Moderate neurodevelopmental impairment (NDI) was evident in 5.7 % of non-BPD infants, 11 % BPD1, 15 % BPD2, 15 % BPD3 infants. Severe NDI was seen in 1.7 % non-BPD infants, 3.4 % BPD1, 7.3 % BPD2, 35 % BPD3 infants. After adjusting for confounders, infants with BPD2 (OR 2.24, 99.9 % CI 1.25 to 5.77) or BPD3 (OR 5.99, 99.9 % CI 1.27 to 46.77) were more likely to have moderate-severe NDI compared to non-BPD infants. Conclusion: The majority of infants with BPD were discharged home without respiratory support and had better neurocognitive outcomes in early childhood compared to those that required home-based oxygen or respiratory support.

PMID: 38490918

#### 21. Neural correlates of bilateral proprioception and adaptation with training

Sebastian Rueda Parra, Joel C Perry, Eric T Wolbrecht, Disha Gupta

PLoS One. 2024 Mar 15;19(3):e0299873. doi: 10.1371/journal.pone.0299873. eCollection 2024.

Bilateral proprioception includes the ability to sense the position and motion of one hand relative to the other, without looking. This sensory ability allows us to perform daily activities seamlessly, and its impairment is observed in various neurological disorders such as cerebral palsy and stroke. It can undergo experience-dependent plasticity, as seen in trained piano players. If its neural correlates were better understood, it would provide a useful assay and target for neurorehabilitation for people with impaired proprioception. We designed a non-invasive electroencephalography-based paradigm to assess the neural features relevant to proprioception, especially focusing on bilateral proprioception, i.e., assessing the limb distance from the body with the other limb. We compared it with a movement-only task, with and without the visibility of the target hand. Additionally, we explored proprioceptive accuracy during the tasks. We tested eleven Controls and nine Skilled musicians to assess whether sensorimotor event-related spectral perturbations in  $\mu$  (8-12Hz) and low- $\beta$  (12-18Hz) rhythms differ in people with musical instrument training, which intrinsically involves a bilateral proprioceptive component, or when new sensor modalities are added to the task. The Skilled group showed significantly reduced μ and low-β suppression in bilateral tasks compared to movement-only, a significative difference relative to Controls. This may be explained by reduced top-down control due to intensive training, despite this, proprioceptive errors were not smaller for this group. Target visibility significantly reduced proprioceptive error in Controls, while no change was observed in the Skilled group. During visual tasks, Controls exhibited significant μ and low-β power reversals, with significant differences relative to proprioceptive-only tasks compared to the Skilled group-possibly due to reduced uncertainty and top-down control. These results provide support for sensorimotor  $\mu$  and low-β suppression as potential neuromarkers for assessing proprioceptive ability. The identification of these features is significant as they could be used to quantify altered proprioceptive neural processing in skill and movement disorders. This in turn can be useful as an assay for pre and post sensory-motor intervention research.

PMID: 38489319

#### 22. Opioid prescription patterns among commercially insured children with and without cerebral palsy

Lubna Ayoubi, Jessica Pruente, Alecia K Daunter, Steven R Erickson, Daniel Whibley, Daniel G Whitney

J Pediatr Rehabil Med. 2024 Mar 14. doi: 10.3233/PRM-230009. Online ahead of print.

Purpose: This study aimed to describe opioid prescription patterns for children with vs. without cerebral palsy (CP). Methods: This cohort study used commercial claims from 01/01/2015-12/31/2016 and included children aged 2-18 years old with and without CP. Opioid prescription patterns (proportion exposed, number of days supplied) were described. A zero-inflated generalized linear model compared the proportion exposed to opioids in the follow-up year (2016) and, among those exposed, the number of days supplied opioids between cohorts before and after adjusting for age, gender, race, U.S. region of residence, and the number of co-occurring neurological/neurodevelopmental disabilities (NDDs). Results: A higher proportion of children with (n = 1,966) vs. without (n = 1,219,399) CP were exposed to opioids (12.1% vs. 5.3%), even among the youngest age group (2-4 years: 9.6% vs. 1.8%), and had a greater number of days supplied (median [interquartile range], 8 [5-13] vs. 6 [4-9] days; P < 0.05). Comparing children with opioid exposure with vs. without CP, a greater number of days supplied was identified for older age, Asian race/ethnicity, and without co-occurring NDDs, and a lower number of days supplied was observed for Black race/ethnicity and with ≥1 co-occurring NDDs. Conclusion: Children with CP are more likely to be exposed to opioids and have a higher number of days supplied.

PMID: 38489199

#### 23. Resilience and adolescence-transition in youth with developmental disabilities and their families: a scoping review

Naomi Zukerman, Emily Bottone, Maya Low, Tatiana Ogourtsova

Review Front Rehabil Sci. 2024 Feb 27:5:1341740. doi: 10.3389/fresc.2024.1341740. eCollection 2024.

Background: Children with neurodevelopmental disabilities (NDDs, e.g., cerebral palsy) and their caregivers face lifelong and impactful challenges, particularly during life-transition periods such as adolescence. One's resilience emerges as an essential ability to navigate this vulnerable phase. Resilience is a complex concept that embeds multiple factors on various levels. Little is known about what resilience factors are pivotal in youth with NDDs and their families as they transition into adolescence and how these are addressed as part of existing targeted interventions. Objectives: This review explored the concept of resilience in youth with NDDs and their families. Specific aims included describing salient resilience factors in adolescents with NDDs and their families and to describe how resilience is addressed as part of targeted interventions. Methods: Using the Arskey and O'Malley framework, six steps were undertaken, including a comprehensive literature search (n = 5 databases), transparent study selection, detailed data extraction with a coding scheme (n = 46 factors), results' collating with numerical and inductive content analysis, and consultation with three key stakeholders. Results: The study screened 1,191 publications, selecting fiftyeight (n = 58; n = 52 observational and n = 6 intervention) studies. Findings revealed that resilience in this context is closely linked to more than forty factors across four levels (individual; family; school/peers; and community). Pivotal factors include social and emotional competence, optimism, and family/peer relationships. While existing interventions targeting resilience show promising results, few programs are available and generalizable to different NDDs. Stakeholders highlighted the importance of addressing resilience factors that are not targeted in existing interventions: caregivers' self-efficacy and selfesteem, as well as youth's and caregiver's confidence. Preferences for and advantages of online delivery for support programs and individual/group features also emerged. Conclusion: The review emphasizes the need for a holistic approach to support youth with NDDs and their families during adolescence transition. To enhance their resilience, recognizing caregivers' roles, customizing interventions, and exploring new implementation formats are avenues that align with the current evidence and opportunities for practical development in this field.

PMID: 38476963

#### 24. Temporal Trends in Severe Brain Injury and Associated Outcomes in Very Preterm Infants

Abdul Razak, Emily Johnston, Alice Stewart, Marissa A T Clark, Penelope Stevens, Margaret Charlton, Flora Wong, C McDonald, Rod W Hunt, Suzanne Miller, Atul Malhotra

Neonatology. 2024 Mar 12:1-10. doi: 10.1159/000537801. Online ahead of print.

Introduction: Severe brain injury (SBI), including severe intraventricular haemorrhage (sIVH) and cystic periventricular leukomalacia, poses significant challenges for preterm infants, yet recent data and trends are limited. Methods: Analyses were conducted using the Australian and New Zealand Neonatal Network data on preterm infants born <32 weeks' gestation admitted at Monash Children's Hospital, Australia, from January 2014 to April 2021. The occurrence and trends of SBI and sIVH among preterm infants, along with the rates and trends of death and neurodevelopmental impairment (NDI) in SBI infants were assessed. Results: Of 1,609 preterm infants, 6.7% had SBI, and 5.6% exhibited sIVH. A total of 37.6% of infants with SBI did not survive to discharge, with 92% of these deaths occurring following redirection of clinical care. Cerebral palsy was diagnosed in 65.2% of SBI survivors, while 86.4% of SBI survivors experienced NDI. No statistically significant

differences were observed in the temporal trends of SBI (adjusted OR [95% CI] 1.08 [0.97-1.20]; p = 0.13) or sIVH (adjusted OR [95% CI] 1.09 [0.97-1.21]; p = 0.11). Similarly, there was no statistically significant difference noted in the temporal trend of the composite outcome, which included death or NDI among infants with SBI (adjusted OR [95% CI] 0.90 [0.53-1.53]; p = 0.71). Conclusion: Neither the rates of SBI nor its associated composite outcome of death or NDI improved over time. A notable proportion of preterm infants with SBI faced redirection of care and subsequent mortality, while most survivors exhibited adverse neurodevelopmental challenges. The development of better therapeutic interventions is imperative to improve outcomes for these vulnerable infants.

PMID: 38471459

#### 25. Neonatal magnesium sulphate for neuroprotection: A systematic review and meta-analysis

Emily Shepherd, Tasneem Karim, Sarah McIntyre, Shona Goldsmith, Amy Keir, Nadia Badawi, Rod W Hunt, Robert Galinsky

Review Dev Med Child Neurol. 2024 Mar 11. doi: 10.1111/dmcn.15899. Online ahead of print.

Aim: To review the evidence of the effects of neonatal magnesium sulphate for neuroprotection in perinatal asphyxia and hypoxic-ischaemic encephalopathy (HIE). Method: This was a systematic review of randomized controlled trials (RCTs) (with meta-analysis) and non-RCTs assessing magnesium sulphate for treating perinatal asphyxia and HIE at 35 weeks or more gestation (primary outcomes: neonatal death and death or long-term major neurodevelopmental disability). Results: Twenty-five RCTs (2099 infants) and four non-RCTs (871 infants) were included, 23 in low- and middle-income countries (LMICs). In RCTs, reductions in neonatal death with magnesium sulphate versus placebo or no treatment (risk ratio [RR] = 0.68; 95% confidence interval [CI] = 0.53-0.86; 13 RCTs), and magnesium sulphate with melatonin versus melatonin alone (RR = 0.74; 95% CI = 0.58-0.95; one RCT) were observed. No difference in neonatal death was seen for magnesium sulphate with therapeutic hypothermia versus therapeutic hypothermia alone (RR = 0.66, 95% CI = 0.34-1.26; three RCTs), or magnesium sulphate versus phenobarbital (RR = 3.00; 95% CI = 0.86-10.46; one RCT). No reduction in death or long-term neurodevelopmental disability (RR = 0.52; 95% CI = 0.14-1.89; one RCT) but reductions in several short-term adverse outcomes were observed with magnesium sulphate. Evidence was low- to very-low certainty because of risk of bias and imprecision. Interpretation: Given the uncertainty of the current evidence, further robust neonatal magnesium sulphate research is justified. This may include high-quality studies to determine stand-alone effects in LMICs and effects with and after therapeutic hypothermia in high-income countries.

PMID: 38468452

## 26. Postoperative Complications of Surgery for Cervical Spondylotic Myelopathy with and Without Athetoid Cerebral Palsy

Naohiro Tachibana, Nobuaki Michihata, Takeshi Oichi, Kosei Nagata, Hideki Nakamoto, Nozomu Ohtomo, Yuichi Yoshida, Koji Nakajima, Junya Miyahara, So Kato, Toru Doi, Yuki Taniguchi, Yoshitaka Matsubayashi, Sakae Tanaka, Hideo Yasunaga, Yasushi Oshima

Global Spine J. 2024 Mar 11:21925682241239610. doi: 10.1177/21925682241239610. Online ahead of print.

Study design: Retrospective cohort study. Objectives: With the aging of the Japanese population, patients with athetoid cerebral palsy (ACP) are getting older, and the rate of surgery for CSM is increasing in ACP patients. However, postoperative complications of such surgery among adult patients with ACP have not been reported yet. We investigated postoperative complications of surgery for CSM with ACP and compared them with those of surgery for CSM without ACP using a national inpatient database of Japan. Methods: Using the Diagnosis Procedure Combination database, we identified 61382 patients who underwent surgery for CSM from July 2010 to March 2018. We examined patient backgrounds, surgical procedures, and type of hospital, and a 4:1 propensity score matching was performed to compare the outcomes between the non-ACP and ACP groups. Results: There were 60 847 patients without ACP and 535 patients with ACP. The mean age was 68.5 years in the non-ACP group and 55 years in the ACP group. The percentages of patients who underwent fusion surgery were 21.6% and 68.8% in the non-ACP and ACP groups, respectively. The 4:1 propensity score matching selected 1858 in the non-ACP group and 465 in the ACP group. The ACP group was more likely to have postoperative urinary tract infection (.4% vs 2.8%, P < .001), postoperative pneumonia (.4% vs 2.4%, P < .001), and 90-day readmission for reoperation (1.9% vs 4.3%, P = .003). Conclusions: We found that ACP patients were more vulnerable to postoperative complications and reoperation after CSM than non-ACP patients.

PMID: 38468399

#### 27. Minimum Effective Dose of Clemastine in a Mouse Model of Preterm White Matter Injury

Elizabeth Odell, Nora Jabassini, Björn Schniedewind, Sarah E Pease-Raissi, Adam Frymoyer, Uwe Christians, Ari J Green, Jonah R Chan, Bridget E L Ostrem

bioRxiv [Preprint]. 2024 Mar 1:2024.02.08.578953. doi: 10.1101/2024.02.08.578953.

Background: Preterm white matter injury (PWMI) is the most common cause of brain injury in premature neonates. PWMI involves a differentiation arrest of oligodendrocytes, the myelinating cells of the central nervous system. Clemastine was previously shown to induce oligodendrocyte differentiation and myelination in mouse models of PWMI at a dose of 10 mg/kg/day. The minimum effective dose (MED) of clemastine is unknown. Identification if the MED is essential for maximizing safety and efficacy in neonatal clinical trials. We hypothesized that the MED in neonatal mice is lower than 10 mg/kg/day. Methods: Mouse pups were exposed to normoxia or hypoxia (10% FiO 2) from postnatal day 3 (P3) through P10. Vehicle or clemastine fumarate at one of four doses (0.5, 2, 7.5 or 10 mg/kg/day) was given orally to hypoxia-exposed pups. At P14, myelination was assessed by immunohistochemistry and electron microscopy to determine the MED. Clemastine pharmacokinetics were evaluated at steady-state on day 8 of treatment. Results: Clemastine rescued hypoxia-induced hypomyelination with a MED of 7.5 mg/kg/day. Pharmacokinetic analysis of the MED revealed C max 44.0 ng/mL, t 1/2 4.6 hours, and AUC 24 280.1 ng\*hr/mL. Conclusion: Based on these results, myelination-promoting exposures should be achievable with oral doses of clemastine in neonates with PWMI.

PMID: 38464078