Cerebral palsy research news

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Interventions and Management

1. Upper Extremity Dystonia Features in People With Spastic Cerebral Palsy

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Neurol Clin Pract. 2023 Dec;13(6):e200207. doi: 10.1212/CPJ.0000000000200207. Epub 2023 Sep 29.

Background and objectives: Dystonia in cerebral palsy (CP) is debilitating and common, but underdiagnosed, especially when coexistent with spasticity. With dedicated research-based assessment, dystonia is found in most people with spastic CP but is only clinically diagnosed in the minority. To begin addressing the high rates of dystonia underdiagnosis in this population, we determined the key feature experts use to assess upper extremity dystonia in people with spastic CP. Methods: In this prospective cohort study, 3 pediatric movement disorder specialists assessed upper extremity dystonia in neurologic examination videos of people with spastic CP and isolated periventricular leukomalacia (PVL) on brain MRI (i.e., those with a brain injury pattern typical for spastic CP). Dystonia severity was rated using the 10-point Global Dystonia Severity Rating Scale, first by each expert independently and then again after consensus-building discussion. Conventional content analysis of these discussions revealed salient features ("codes") that experts used to assess upper extremity dystonia. Code frequency distributions were compared between dystonia severity categories using $\chi 2$ tests. Results: We identified 96 people with spastic CP with isolated PVL on brain MRI seen in the St. Louis Children's Hospital CP Center between 2005 and 2018. Of them, 26 people were able and willing to be recorded while doing a standardized set of upper extremity examination maneuvers (age 4-25 years; 28% nonambulatory, 77% White). When assessing their videos, experts cited the "hand" less often and "shoulder" more often with increasing dystonia severity ($p < 0.005$, $\chi 2$ test). "Mirror movements" and the "hand open/close" examination maneuver were cited significantly more frequently in videos when experts were attempting to distinguish between no dystonia and mild dystonia ($p < 0.005$). Discussion: Expert clinicians use distinct movement features to assess upper extremity dystonia in people with spastic CP and PVL. Attention to involuntary shoulder (vs hand) movements can help gauge dystonia severity. Differentiation between mirror movements and dystonia, particularly during the hand open/close examination maneuver, may help identify mild dystonia. These results can help guide upper extremity dystonia assessment in people with spastic CP, thus potentially helping mitigate dystonia underdiagnosis.

PMID: 37780812

2. Prevention of hip dislocation in severe cerebral palsy (GMFCS III-IV-V): an interdisciplinary and multi-professional Care Pathway for clinical best practice implementation

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Eur J Phys Rehabil Med. 2023 Oct 5. doi: 10.23736/S1973-9087.23.07978-9. Online ahead of print.

Background: Hip displacement (HD) and dislocation in severe Cerebral Palsy (CP) (GMFCS III, IV, V) are important causes of worsening disability and quality of life. Prevention must be started from the first months of life through screening programs and early treatments, both conservative and surgical. Evidence from Clinical Practice Guidelines also suggests the

development of Care Pathways for good clinical practice. At the beginning of 2020 an interdisciplinary, multi-professional working group, composed of 26 members (including Physiatrists, Physiotherapist, Neuro-psychomotor Therapists and Orthopedists representing the respective Italian Scientific Societies) with the involvement of the FightTheStroke Foundation families' association, was set up. Aim: The aim of the multi-professional panel was the production of evidence-based recommendations for the Care Pathway "Prevention of Hip Displacement in children and adolescents with severe CP" for best clinical practice implementation in our national context. Design: Clinical Care Pathway (Clinical Practice Guideline). Setting: Inpatient and outpatient. Population: Children with severe CP (GMFCS III-IV-V). Methods: The recommendations of this Care Pathway were developed using the American Academy for Cerebral Palsy and Developmental Medicine (AACPDM) guidelines for Care Pathways development and the Grading of Recommendations Assessment Development and Evaluation (GRADE ADOLOPMENT) working group for adoption or adaption or de novo development of recommendations from high-quality guidelines. In 2020 a multidisciplinary working group (WG) developed four research questions on the prevention of HD on the following topics: screening, botulinum toxin treatment, postural management and preventive soft tissue surgery. A comprehensive review of the biomedical literature was performed on each question. Guidelines, Systematic Reviews and Primary studies were retrieved through a top-down approach. References were screened according to inclusion criteria and quality was assessed by means of specific tools. A list of recommendations was then produced divided by intervention (screening programs, postural management, botulinum toxin, preventive surgery). In a series of meetings, the panel graduated recommendations using the GRADE evidence to decision frameworks. Results: Fifteen recommendations were developed: seven on screening programs, four on postural management strategies, one on botulinum toxin, and three on preventive surgery. Evidence quality was variable (from very low to moderate) and only a few strong recommendations were made. Conclusions: In severe CP at high risk of hip dislocation, it is strongly recommended to start early hip surveillance programs. In our national context, there is a need to implement Screening programs and dedicated Network teams. We also strongly recommend a comprehensive approach shared with the families and goal-oriented by integrating the different therapeutic interventions, both conservative and not, within Screening programs. Clinical rehabilitation impact: Implementing a comprehensive multi-professional approach for the prevention of hip dislocation in severe CP.

PMID: 37796120

3. Hip reconstruction in cerebral palsy: Lessons from a single center and 137 hips

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J Child Orthop. 2023 Sep 5;17(5):469-480. doi: 10.1177/18632521231196846. eCollection 2023 Oct.

Background: This large, retrospective, single-center study aimed to compare the outcomes of unilateral hip reconstruction and bilateral hip reconstruction in children with non-ambulatory cerebral palsy and ascertain risk factors for recurrent instability and reoperation. Method: We performed a retrospective review of 137 hip reconstructions performed for patients with cerebral palsy. Preoperative and postoperative clinical and radiological parameters were documented, including hip migration percentage, acetabular index, the Gross Motor Function Classification System, the Melbourne Cerebral Palsy Hip Classification System, hip abduction, and pelvic obliquity. Results: Overall, 49 patients underwent bilateral hip reconstruction, and 37 patients underwent unilateral hip reconstruction. In the unilateral hip reconstruction group, the reconstructed hip remained stable (with a migration percentage < 33%) in 59% of patients compared to 74.4% of the more affected hips in the bilateral hip reconstruction group ($p = 0.02$). Of the unreconstructed hip in the unilateral hip reconstruction group, 74.4% remained stable (with a migration percentage < 33%), compared to 78.8% of the less affected hips in the bilateral hip reconstruction group. A level pelvis was maintained at final follow-up in significantly more patients in the bilateral hip reconstruction group than the unilateral hip reconstruction group ($p = 0.002$). Further surgical intervention was performed in 41% of the unilateral hip reconstruction group, compared with 11.5% in the bilateral hip reconstruction group ($p = 0.001$). Surgery performed under the age of 8 years and not performing an acetabular osteotomy were found to be risk factors for recurrent instability in all groups. Conclusion: Our series suggests that in terms of recurrent instability after hip reconstruction in cerebral palsy, protective factors against this complication include bilateral hip reconstruction, hip reconstruction after 8 years of age, and the use of an acetabular osteotomy.

PMID: 37799318

4. Longitudinal trajectory of medial gastrocnemius muscle growth in the first years of life

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Dev Med Child Neurol. 2023 Oct 3. doi: 10.1111/dmcn.15763. Online ahead of print.

Aim: To define the longitudinal trajectory of gastrocnemius muscle growth in 6- to 36-month-old children with and without spastic cerebral palsy (SCP) and to compare trajectories by levels of gross motor function (Gross Motor Function Classification System, GMFCS) and presumed brain-lesion timing. Method: Twenty typically developing children and 24 children with SCP (GMFCS levels I-II/III-IV = 15/9), were included (28/16 females/males; mean age at first scan 15.4 months [standard deviation 4.93, range 6.24-23.8]). Three-dimensional freehand ultrasound was used to repeatedly assess muscle volume, length, and cross

-sectional area (CSA), resulting in 138 assessments (mean interval 7.9 months). Brain lesion timing was evaluated with magnetic resonance imaging classification. Linear mixed-effects models defined growth rates, adjusted for GMFCS levels and presumed brain-lesion timing. Results: At age 12 months, children with SCP showed smaller morphological muscle size than typically developing children (5.8 mL vs 9.8 mL, $p < 0.001$), while subsequently no differences in muscle growth were found between children with and without SCP (muscle volume: 0.65 mL/month vs 0.74 mL/month). However, muscle volume and CSA growth rates were lower in children classified in GMFCS levels III and IV than typically developing children and those classified in GMFCS levels I and II, with differences ranging from -56% to -70% ($p < 0.001$). Interpretation: Muscle growth is already hampered during infancy in SCP. Muscle size growth further reduces with decreasing functional levels, independently from the brain lesion. Early monitoring of muscle growth combined with early intervention is needed.

PMID: 37786988

5. Can developmental trajectories in gait variability provide prognostic clues in motor adaptation among children with mild cerebral palsy? A retrospective observational cohort study

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Front Hum Neurosci. 2023 Sep 19:17:1205969. doi: 10.3389/fnhum.2023.1205969. eCollection 2023.

Aim: To investigate whether multiple domains of gait variability change during motor maturation and if this change over time could differentiate children with a typical development (TDC) from those with cerebral palsy (CwCP). Methods: This cross-sectional retrospective study included 42 TDC and 129 CwCP, of which 99 and 30 exhibited GMFCS level I and II, respectively. Participants underwent barefoot 3D gait analysis. Age and parameters of gait variability (coefficient of variation of stride-time, stride length, single limb support time, walking speed, and cadence; as well as meanSD for hip flexion, knee flexion, and ankle dorsiflexion) were used to fit linear models, where the slope of the models could differ between groups to test the hypotheses. Results: Motor-developmental trajectories of gait variability were able to distinguish between TDC and CwCP for all parameters, except the variability of joint angles. CwCP with GMFCS II also showed significantly higher levels of gait variability compared to those with GMFCS I, these levels were maintained across different ages. Interpretation: This study showed the potential of gait variability to identify and detect the motor characteristics of high functioning CwCP. In future, such trajectories could provide functional biomarkers for identifying children with mild movement related disorders and support the management of expectations.

PMID: 37795211

6. Gait analysis patterns and rehabilitative interventions to improve gait in persons with hereditary spastic paraplegia: a systematic review and meta-analysis

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Front Neurol. 2023 Sep 20:14:1256392. doi: 10.3389/fneur.2023.1256392. eCollection 2023.
Background: Hereditary spastic paraplegias (HSPs) are a group of inheritance diseases resulting in gait abnormalities, which may be detected using instrumented gait analysis. The aim of this systematic review was 2-fold: to identify specific gait analysis patterns and interventions improving gait in HSP subjects. Methods: A systematic review was conducted in PubMed, Cochrane Library, REHABDATA, and PEDro databases, in accordance with reporting guidelines of PRISMA statement and Cochrane's recommendation. The review protocol was recorded on the PROSPERO register. Patients with pure and complicated HSP of any age were included. All types of studies were included. Risk of bias, quality assessment, and meta-analysis were performed. Results: Forty-two studies were included: 19 were related to gait analysis patterns, and 24 were intervention studies. The latter ones were limited to adults. HSP gait patterns were similar to cerebral palsy in younger subjects and stroke in adults. Knee hyperextension, reduced range of motion at knee, ankle, and hip, reduced foot lift, and increased rapid trunk and arm movements were reported. Botulinum injections reduced spasticity but uncovered weakness and improved gait velocity at follow-up. Weak evidence supported intrathecal baclofen, active intensive physical therapy (i.e., robot-assisted gait training, functional exercises, and hydrotherapy), and functional electrical stimulation. Some improvements but adverse events were reported after transcranial magnetic stimulation, transcutaneous spinal direct current stimulation, and spinal cord stimulation implant. Conclusion: Knee hyperextension, non-sagittal pelvic movements, and reduced ROM at the knee, ankle, and hip represent the most peculiar patterns in HSP, compared to diplegic cerebral palsy and stroke. Botulinum improved comfortable gait velocity after 2 months. Nonetheless, interventions reducing spasticity might result in ineffective functional outcomes unveiling weakness. Intensive active physical therapy and FES might improve gait velocity in the very short term.

PMID: 37799279

7. Does Botulinum Toxin Injection Exacerbate Sarcopenia and Bone Mass in Individuals With Cerebral Palsy?

Yu-Chi Su, Meng-Che Tsai, Chung-Ying Lin, Jen Yang, Pei-Shan Wu, Hsiu-Ching Yang, Yu-Ching Lin

Pediatr Neurol. 2023 Sep 7:149:32-38. doi: 10.1016/j.pediatrneurol.2023.09.002. Online ahead of print.

Background: Botulinum toxin (BoNT) causes sarcopenia and low bone mass in animal studies. Whether such effect exists in children and adolescents with spastic cerebral palsy (CP) is not clear yet. To investigate the influences of BoNT on grip strength (GS), skeletal muscle mass, and bone mineral density (BMD) in children and adolescents with spastic CP, we conducted this uncontrolled longitudinal study. Methods: The body composition of individuals with spastic CP were measured by dual-energy X-ray absorptiometry at preinjection and at 12 and 24 weeks after BoNT intervention. Sarcopenia was defined as meeting both decreased GS and low muscle mass. Twenty-five participants were enrolled (mean age 8.5 years). Results: Before BoNT intervention, four adolescents had sarcopenia and low bone mass. When the body composition was analyzed as four limbs, trunk, and head, the skeletal muscle mass of the injected limbs, appendicular skeletal muscle mass, and total body less head BMD increased significantly over 24-week follow-up period (P = 0.0117, 0.0032, 0.0229), whereas the GS remained unchanged. When the body composition was analyzed as segments derived from bilateral arms, forearms, hands, thighs, and lower legs, the skeletal muscle mass (P = 0.0113) but not BMD of the injected segments increased significantly over the 24 weeks. The prevalence of low muscle mass, decreased GS, sarcopenia, and low bone mass did not change over 24 weeks. Conclusions: The present study showed that BoNT does not exacerbate sarcopenia and low bone mass in individuals with spastic CP.

PMID: 37776658

8. The effectiveness of dual task exercise training on balance, mobility, physical performance, and quality of life in children with cerebral palsy: a single-blind randomized controlled trial

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Ir J Med Sci. 2023 Sep 30. doi: 10.1007/s11845-023-03530-3. Online ahead of print.

Background: Most activities of daily living require more than one activity, including motor-motor or motor-cognitive task. Few studies have investigated the effects of dual-task training in children with cerebral palsy (CP). Aims: This study was aimed at investigating the effectiveness of motor-cognitive dual-task exercise training in children with cerebral palsy (CP). Methods: A single-blind randomized controlled trial was conducted with thirty children with CP. Participants were randomized into dual group training group (DTG) and control group (CG). Children were evaluated before the intervention and after 12 weeks of treatment with Gross Motor Function Classification Scale (GMFCS), Pediatric Berg Balance Test (PBBS), Single Leg Stance Test (SLST), Timed Up and Go Test (TUG), 3 Meter Backwards Walk Test (3-MBWT), 6 Meter Walk Test (6-MWT), and General Children's Quality of Life Measure (KINDL). Results: DTG demonstrated significantly improved all KINDL scores ($p < 0.01$). In comparisons between groups adjusted analysis, results showed a better improvement of all KINDL scores in-favor-of DTG ($p < 0.01$). Significant improvements were found in all SLST scores and PBS in DTG ($p < 0.01$). Adjusted analysis results proved there was a significant improvement in all balance scores on behalf of DTG ($p < 0.01$). Significant improvement was observed in DTG for TUG, 3MBWT, and 6MWT scores ($p = 0.001$). An improvement in-favor-of DTG was found for all performance tests in the adjusted analysis ($p < 0.001$). Conclusion: The results of this randomized controlled trial highlighted the advantage of dual-task training on balance, physical performance, and quality of life in children with CP.

PMID: 37777679

9. Participate CP 2: optimising participation in physically active leisure for children with cerebral palsy - protocol for a phase III randomised controlled trial

Leanne Sakzewski, Sarah Elizabeth Reedman, Catherine Elliott, Jenny Ziviani, Iona Novak, Stewart Trost, Annette Majnemer, Tracy Comans, Keiko Shikako, Robert S Ware, Lynda McNamara, Sian Williams, Syed Afroz Keramat, Denise Brookes, Roslyn N Boyd

BMJ Open. 2023 Oct 3;13(10):e075570. doi: 10.1136/bmjopen-2023-075570.

Introduction: Children with cerebral palsy (CP) participate less in physical activities and have increased sedentary behaviour compared with typically developing peers. Participate CP is a participation-focused therapy intervention for children with CP with demonstrated efficacy in a phase II randomised controlled trial (RCT) to increase perceived performance of physical activity participation goals. This study will test the effectiveness of Participate CP in a multisite phase III RCT. Methods and analysis: One hundred children with CP, aged 8-14 years, classified Gross Motor Function Classification System levels I-IV will be randomised to either (1) receive Participate CP once/week for 1 hour for 12 weeks, or (2) waitlist control, usual care group. The waitlist group will then receive Participate CP following the 26-week retention time point. Outcomes will be assessed at baseline, 12 weeks and then 26 weeks post baseline. The primary outcomes are (1) self-reported participation goal performance on the Canadian Occupational Performance Measure at 12 weeks and (2) daily time in moderate-to-vigorous physical activity. Secondary outcomes include home and community participation frequency, involvement and environmental supportiveness, contextual barriers to participation, quality of life, intrinsic motivation for physical activities, child perception of an autonomy-supportive climate for physical activities and physical literacy at 12 and 26 weeks post study entry. Ethics and dissemination: The Children's Health Queensland Hospital and Health Service, The University of Queensland and the New

Zealand Health and Disability Ethics Committees have approved this study. Findings will be disseminated in peer-reviewed journals and conference presentations. Trial registration number: ACTRN12618000206224.

PMID: 37788925

10. Using artificial intelligence-based technologies to detect clinically relevant changes of gross motor function in children with cerebral palsy

Leonie Schafmeyer, Heike Losch, Christiane Bossier, Isabel Lanz, Heidrun Lioba Wunram, Eckhard Schoenau, Ibrahim Duran

Dev Med Child Neurol. 2023 Oct 4. doi: 10.1111/dmcn.15744. Online ahead of print.

Aim: To compare the 66-item Gross Motor Function Measure (GMFM-66) with the reduced version of the GMFM-66 (rGMFM-66) with respect to the detection of clinically relevant changes in gross motor function in children with cerebral palsy (CP). Method: The study was a retrospective single centre analysis of children with CP who participated in a rehabilitation programme. Overall, 1352 pairs of GMFM-66 and rGMFM66 measurements with a time interval of 5 to 7 months were available. To measure clinically relevant changes in gross motor function, the individual effect size (iES) was calculated. Results: The study population consisted of 1352 children (539 females), mean age 6 years 4 months (SD 2 years 4 months). The iES based on the GMFM-66 and the rGMFM-66 showed a significant correlation ($r = 0.84$, $p < 0.001$). The analysis of the area under the receiver operating characteristic curve showed an excellent agreement for clinically relevant gross motor improvement (Cohen's $d \geq 0.5$; area under the curve = 0.90 [95% confidence interval 0.88-0.92]) or deterioration (Cohen's $d \leq -0.5$; area under the curve = 0.95 [95% confidence interval 0.92-0.97]). Interpretation: Performing the rGMFM-66 saves time compared to the full GMFM-66. The rGMFM-66 showed good agreement with the GMFM-66 with respect to the detection of clinically relevant changes in gross motor function in children with CP, so its use in everyday clinical practice seems justifiable.

PMID: 37794634

11. Strategies supporting parent-delivered rehabilitation exercises to improve motor function after paediatric traumatic brain injury: A systematic review

David Young, Sarah Cawood, Kathryn Mares, Robbie Duschinsky, Wendy Hardeman

Review Dev Med Child Neurol. 2023 Oct 4. doi: 10.1111/dmcn.15773. Online ahead of print.

Aim: To identify and analyse ways in which parents are supported to deliver rehabilitation exercises to their child after traumatic brain injury (TBI), conceptualized as strategies. Method: A systematic search was completed using seven online databases and three grey literature databases, from inception to November 2021. The included studies focused on physical rehabilitation in children after TBI with the involvement of parents as hands-on deliverers or facilitators of rehabilitation (e.g. supervising the exercise). Intervention descriptions were reviewed to identify strategies; this was followed by fine-grained analysis using the Behaviour Change Wheel to identify intervention components. Risk of bias was analysed using the revised Cochrane Risk-of-Bias Tool for Randomized Trials or the Risk Of Bias In Non-randomized Studies - of Interventions. Results: Six interventions including 211 participants and one trial protocol met the inclusion criteria. All studies included a proportion of children diagnosed with TBI and four studies included mixed samples of acquired brain injury or cerebral palsy. All interventions included elements of goal setting and instruction. Interpretation: Interventions focus heavily on the initiation of physical rehabilitation, but focus less on the longer-term maintenance of rehabilitation delivery. Further research should integrate perspectives from parents to inform the development of new interventions.

PMID: 37794644

12. [Re-evaluation of systematic reviews of acupuncture and moxibustion for children with cerebral palsy] [Article in Chinese & Abstract in English, Chinese]

Xiao-Fei Lu, Ya-Wen Tao, Fan Liu, Yu-Qin Xu, Ming-Qiang Gong, Zhuo-Xin Yang

Zhongguo Zhen Jiu. 2023 Oct 12;43(10):1209-16. doi: 10.13703/j.0255-2930.20221008-0004.

Objective: To assess the methodological quality, report quality and evidence quality of the Meta-analysis and systematic reviews of acupuncture and moxibustion for children with cerebral palsy, aiming to provide decision-making basis for clinical treatment. Methods: The systematic reviews and Meta-analysis of acupuncture and moxibustion for children with cerebral palsy were searched in CNKI, Wanfang, VIP, SinoMed, Cochrane Library, PubMed and EMbase. The retrieval time was from the database establishment to June 30th, 2022. AMSTAR 2 (a measurement tool to assess systematic reviews) was used to evaluate the methodological quality， and PRISMA (preferred reporting items for systematic reviews and Meta-analyses) was used to evaluate the report quality, and GRADE was used to evaluate the quality of evidence. Results: A total of 14 systematic

reviews were included, including 37 primary outcome indexes. According to AMSTAR 2 evaluation results, there were 4 low quality studies, 10 very low quality studies, and low scores on items 2, 4, 7, 10 and 16. PRISMA scores ranged from 15 to 25, and the main reporting problems reflected in structured abstracts, program and registration, retrieval, and funding sources, etc. According to the GRADE classification results, there were 3 high quality evidences, 7 medium quality evidences, 10 low quality evidences and 17 very low quality evidences. The main downgrading factors were limitations, imprecision and publication bias. Conclusion: Acupuncture and moxibustion has a certain effect for cerebral palsy in children, but the quality of methodology, reporting and evidence in the included literature is poor, and the comparison of curative effect between different acupuncture and moxibustion methods is unclear.

PMID: 37802530

13. [Properties of the CP-QOL-PCQ to measure health-related quality of life in children and adolescents with cerebral palsy in Argentina] [Article in Spanish & Abstract in English, Spanish]

Silvina Berra, Natalia Herrera Sterren, Javier Sánchez Rosas

Rev Fac Cien Med Univ Nac Cordoba. 2023 Sep 29;80(3):188-204. doi: 10.31053/1853.0605.v80.n3.40042.

Introduction: The impact of cerebral palsy (CP) on quality of life requires the incorporation of subjective assessments of health, functional status, and well-being. A specific health-related quality of life (HRQoL) measure for CP is the Australian CP-QOL questionnaire, whose version for caregivers (PCQ) was adapted in Argentina. This study aimed to achieve the final structure and evaluate the reliability and validity of the CP-QOL-PCQ for the Argentine child and adolescent population. Methods: 100 caregivers of people from 3 to 24 years old with a diagnosis of CP from Argentina participated. In addition to the CP-QOL-PCQ, the KIDSCREEN-27 questionnaire was applied. Analysis of response distributions of all items, inter-item correlations, and internal consistency of the scales were performed using Cronbach's alpha. The convergent validity of the instrument was tested through the correlation with scales of similar content from the KIDSCREEN and through a priori hypotheses in groups of different ages and functional impairment. Results: A final structure of the CP-QOL-PCQ was achieved with 8 multidimensional scales, with satisfactory inter-item correlations (>0.30) and internal consistency (>0.70). Moderate and high correlations (r>0.30) were obtained between similar dimensions of the CP-QOL-PCQ and the KIDSCREEN with similar concepts. Higher HRQoL scores were confirmed at a younger age and at a lower level of functional impairment (Cohen's d >0.20). Conclusion: This provides evidence of reliability and validity to be expected for a HRQoL measurement instrument that can be implemented in the Argentine child and adolescent population with CP.

PMID: 37773342

14. Asymmetric involvement of hands: Psychometric properties of the Turkish version of the Bimanual Fine Motor Function 2.0 classification in children with cerebral palsy

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J Hand Ther. 2023 Sep 28:S0894-1130(23)00119-9. doi: 10.1016/j.jht.2023.08.006. Online ahead of print.

Background: Manual functions affect more than a half of children with Cerebral palsy (CP). Asymmetric involvement of hands may also affect unilateral and bilateral activities of daily life. The Bimanual Fine Motor Function version 2.0 (BFMF-2.0) is a unique functional classification that categorizes the capacity of each hand (what the child can do) during bimanual functions. Purpose: The aim of this study was to investigate the validity and reliability of the Turkish version of the BFMF-2.0 in children with CP. Study design: Clinical measurement and cross-sectional study. Methods: The study included 91 children with CP (56 girls, mean age; 7.41 ± 4.23 years [4-18 years]) and their parents. The Manual Ability Classification System (MACS), the Quality of Upper Extremity Skills Test (QUEST), and the Box and Block Test (BBT) were used for construct and concurrent validity. Experienced/inexperienced therapists and parents classified fine motor capacities of the children via live or video-based observation to assess inter-rater reliability. Three weeks later, the children were reclassified for intra-rater reliability. Results: The Turkish version of the BFMF-2.0 classification was strongly correlated with the MACS (rho = -0.88, $p < 0.001$), the QUEST (rho = 0.80, $p < 0.001$), and the BBT (rho = -0.77, $p < 0.001$). The inter-rater reliability scores were weak to excellent between the parents and the therapists (via live observation, $\kappa w = 0.57$) and also between experienced/inexperienced therapists (via live or video-based observation, $\kappa w = 0.66$-0.79). Intra-rater reliability scores were good to excellent (Intraclass Correlation Coefficient [ICC] = 0.87-0.95). Conclusions: The Turkish version of the BFMF-2.0 classification is valid and reliable and could be applied by experienced and inexperienced therapists via live or video-based observation and by parents via live observation.

PMID: 37777439

15. Two-year neurodevelopmental data for preterm infants born over an 11-year period in England and Wales, 2008-2018: a retrospective study using the National Neonatal Research Database

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Arch Dis Child Fetal Neonatal Ed. 2023 Oct 3:fetalneonatal-2023-325746. doi: 10.1136/archdischild-2023-325746. Online ahead of print.

Objective: United Kingdom guidelines recommend all infants born <30 weeks' gestation receive neurodevelopmental follow-up at 2 years corrected age. In this study, we describe completeness and results of 2-year neurodevelopmental records in the National Neonatal Research Database (NNRD). Design: This retrospective cohort study uses data from the NNRD, which holds data on all neonatal admissions in England and Wales, including 2year follow-up status. Patients: We included all preterm infants born <30 weeks' gestation between 1 January 2008 and 31 December 2018 in England and Wales, who survived to discharge from neonatal care. Main outcome measures: Presence of a 2-year neurodevelopmental assessment record in the NNRD, use of standardised assessment tools, results of functional 2-year neurodevelopmental assessments (visual, auditory, neuromotor, communication, overall development). Results: Of the 41 505 infants included, 24 125 (58%) had a 2-year neurodevelopmental assessment recorded. This improved over time, from 32% to 71% for births in 2008 and 2018, respectively.Of those with available data: 0.4% were blind; 1% had a hearing impairment not correctable with aids; 13% had <5 meaningful words, vocalisations or signs; 8% could not walk without assistance and 9% had severe (≥12 months) developmental delay. Conclusions: The proportion of infants admitted to neonatal units in England and Wales with a 2-year neurodevelopmental record has improved over time. Rates of follow-up data from recent years are comparable to those of bespoke observational studies. With continual improvement in data completeness, the potential for use of NNRD as a source of longer-term outcome data can be realised.

PMID: 37788897

16. Reduced brainstem volume is associated with mobility impairments in youth with cerebral palsy

Michael P Trevarrow, Saihari S Dukkipati, Sarah E Baker, Tony W Wilson, Max J Kurz

J Clin Neurosci. 2023 Oct 4:117:114-119. doi: 10.1016/j.jocn.2023.09.025. Online ahead of print.

Background: Persons with cerebral palsy (CP) have impaired mobility that has been attributed to changes in structure and function within the nervous system. The brainstem is a region that plays a critical role in mobility by connecting the cortex and cerebellum to the spinal cord, yet this region has been largely unstudied in persons with CP. Research question: We used high-resolution structural MRI and biomechanical analyses to examine whether the volume of the whole brainstem and its constituent elements are altered in CP and if these alterations relate to the mobility impairments within this population. Methods: A cohort study was conducted to assess the volume of the whole brainstem, pons, midbrain, medulla, and superior cerebellar peduncle in a cohort of persons with CP (N = 26; Age = 16.3 ± 1.0 years; GMFCS levels I-IV, Females = 12) and a cohort of neurotypical (NT) controls (N = 38; Age = 14.3 ± 0.4 years, Females = 14) using structural MR imaging of the brainstem. Outside the scanner, a digital mat was used to quantify the spatiotemporal gait biomechanics of these individuals. Results: We found a significant decrease in volume of the total brainstem, midbrain, and pons in persons with CP in comparison to the NT controls. Furthermore, we found that the altered volumes were related to reduced gait velocity and step length. Significance: The structural changes in the brainstems of persons with CP may contribute to the mobility impairments that are ubiquitous within this population.

PMID: 37801875

17. Comparison of Outcomes following Surgical Intervention and Inpatient Rehabilitation Stays in Children with Cerebral Palsy

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Introduction: Literature is limited on functional outcomes in children with cerebral palsy (CP) following surgical procedures and a subsequent inpatient rehabilitation unit (IRU) stay. Objective: To compare functional outcomes and length of stay (LOS) in children with CP following a surgical procedure and IRU stay based on the surgical procedure performed, pattern of involvement, etiology, and Gross Motor Function Classification System (GMFCS) level. Design: Retrospective cohort study. Setting: Tertiary care pediatrics. Participants: Pediatric patients with CP who underwent one of three surgical procedures followed by an IRU stay. Interventions: Either selective dorsal rhizotomy (SDR), single-event multilevel orthopedic surgery (SEMLS), or intrathecal baclofen (ITB) pump implantation and subsequent IRU stay. Main outcome measures: IRU LOS, WeeFIM™ total score, sub-scores, and efficiency. Results: Children undergoing SDR had a longer LOS ($p \leq .015$). Children with spastic diplegia, GMFCS level II and prematurity-based CP had higher WeeFIM™ efficiency scores ($p \leq .046$, $\leq .021$,

.034 respectively). Greater changes in WeeFIM™ scores were associated with spastic diplegia, SDR, GMFCS level II, longer LOS, and higher admission scores ($p \leq .045$). Conclusions: While statistically and functionally significant improvements in children with CP following surgical interventions and an IRU stay were seen, those with higher WeeFIM™ change scores tended to have spastic diplegia, undergone SDR, GMFCS level II, longer LOS, and higher admission scores.

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18. Early developmental support for preterm infants based on exploratory behaviors: A parallel randomized controlled study

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Introduction: Preterm infants are at high risk for developmental disabilities, and their parents are at increased risk for high stress. Early intervention programs are applied to reduce these adverse outcomes. The primary aim is to compare the efficacy of the novel Explorer Baby early intervention program for the holistic development of preterm infants. The second objective was to compare the stress levels of their mothers. Methods: Randomized clinical trial with 38 weeks-6 months corrected age preterm infants at low risk for cerebral palsy, randomly assigned to experimental (Explorer Baby) or active control neurodevelopmental therapy (NDT) groups. Fifty-seven infants were enrolled in the study, and 51 (26 Explorer Baby, 25 NDT) completed it. Bayley III was used as a primary outcome before, during, and after the intervention. Results: When we compared the changes between the groups before and after therapy, no significant differences were found in any of the primary or secondary outcomes (between-group comparisons). When comparing the changes in both groups before and after therapy (in-group comparison), the Explorer Baby group demonstrated significant improvements in cognitive (Hedges' g = .83) and explorative language skills (Hedges' g = .65), whereas the NDT group showed improved parent-child dysfunctional interaction (Hedges' g = 2.66) between T0-T1 and T0-T2. Conclusions: The Explorer Baby early intervention program may be a preferred option to support premature infants without brain injury, as it shows greater skill acquisition than NDT, although not statistically significant. Both methods are safe as they support premature babies without negatively affecting mothers' overall stress levels.

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19. Neonatal Hypoxic-Ischemic Encephalopathy and Hypothermia Treatment

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Neonatal hypoxic-ischemic encephalopathy (HIE) is an important clinical entity because it is associated with death and long-term disability, including cognitive impairment, cerebral palsy, seizures, and neurosensory deficits. Over the past 40 years, there has been an intensive search to identify therapies to improve the prognosis of neonates with HIE. Hypothermia treatment represents the culmination of laboratory investigations including small and large animal studies, followed by pilot human studies, and, finally, randomized controlled trials to establish efficacy and safety. Clinical trials have demonstrated that hypothermia treatment reduces mortality and improves early childhood outcome among survivors. Hypoxic-ischemic encephalopathy is a multi-system disease process that requires intensive medical support for brain monitoring and monitoring of non-central nervous system organ dysfunction. Treatment must be conducted in a level III or IV neonatal intensive care unit with infrastructure for an integrated approach to care for critically ill neonates. Hypothermia treatment is the first and currently the only therapy to improve outcomes for neonates with HIE and indicates that HIE is modifiable. However, outcomes likely can be improved further. Hypothermia treatment has accelerated investigation of other therapies to combine with hypothermia. It has also stimulated a more intensive approach to brain monitoring, which allows earlier intervention for complications. Finally, HIE and hypothermia treatment negatively influences the psychological state of affected families, and there is growing recognition of the importance of trauma-informed principles to guide medical professionals.

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20. SIGnature Libraries: A roadmap for the formation of special interest group libraries

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Objective: "SIGnature Libraries" channel the dynamism of academic society-based special interest groups (SIG) to systematically identify and provide user-oriented access to essential literature for a subspecialty field in a manner that keeps pace with the field's continuing evolution. The libraries include literature beyond clinical trial data to encompass historical context, diagnostic conceptualization, and community organization materials to foster a holistic understanding of how neurologic conditions affect individuals, their community, and their lived experience. Methods: Utilizing a modified-Delphi approach, Child Neurology Society's Cerebral Palsy (CP) SIG (n = 75) administered two rounds of literature submissions and ratings. A final review by an 11-member international advisory group determined threshold ratings for resource inclusion and the library's final structure. Results: Seventy-nine articles were submitted for the first Delphi round and 22 articles for the second Delphi round. Survey response rates among SIG members were 29/75 for the first round and 24/75 for the second round. The advisory board added additional articles in the final review process in view of the overall project goal. A total of 60 articles were included in the final library, and articles were divided into seven sections and stratified by rating scores. A process for ongoing revisions of the library was determined. The library will be published on the Child Neurology Society website and made publicly accessible. Conclusions: The CP SIGnature Library offers learners an unprecedented resource that provides equitable access to latest consensus guidelines, existing seminal datasets, up-to-date review articles, and other patient care tools. A distinctive feature of the library is its intentional large scope and depth, presented in a stratified fashion relative to the consensus-determined importance of each article. Learners can efficiently navigate the library based on individual interests and goals, and the library can be used as core curriculum for CP education.

PMID: 37795255

21. Caffeine versus other methylxanthines for the prevention and treatment of apnea in preterm infants

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Background: Methylxanthines, including caffeine, theophylline, and aminophylline, work as stimulants of the respiratory drive, and decrease apnea of prematurity, a developmental disorder common in preterm infants. In particular, caffeine has been reported to improve important clinical outcomes, including bronchopulmonary dysplasia (BPD) and neurodevelopmental disability. However, there is uncertainty regarding the efficacy of caffeine compared to other methylxanthines. Objectives: To assess the effects of caffeine compared to aminophylline or theophylline in preterm infants at risk of apnea, with apnea, or in the peri-extubation phase. Search methods: We searched CENTRAL, MEDLINE, Embase, Epistemonikos, the World Health Organization (WHO) International Clinical Trials Registry Platform (ICTRP), and clinicaltrials.gov in February 2023. We also checked the reference lists of relevant articles to identify additional studies. Selection criteria: Studies: randomized controlled trials (RCTs) and quasi-RCTs Participants: infants born before 34 weeks of gestation for prevention and extubation trials, and infants born before 37 weeks of gestation for treatment trials Intervention and comparison: caffeine versus theophylline or caffeine versus aminophylline. We included all doses and duration of treatment. Data collection and analysis: We used standard methodological procedures expected by Cochrane. We evaluated treatment effects using a fixed-effect model with risk ratio (RR), risk difference (RD), and 95% confidence intervals (CI) for categorical data, and mean, standard deviation, and mean difference for continuous data. We used the GRADE approach to evaluate the certainty of evidence. Main results: We included 22 trials enrolling 1776 preterm infants. The indication for treatment was prevention of apnea in three studies, treatment of apnea in 13 studies, and extubation management in three studies. In three studies, there were multiple indications for treatment, and in one study, the indication for treatment was unclear. In 19 included studies, the infants had a mean gestational age between 28 and 32 weeks and a mean birth weight between 1000 g and 1500 g. One study's participants had a mean gestational age of more than 32 weeks, and two studies had participants with a mean birth weight of 1500 g or more. Caffeine administrated for any indication may result in little to no difference in all-cause mortality prior to hospital discharge compared to other methylxanthines (RR 1.12, 95% CI 0.68 to 1.84; RD 0.02, 95% CI -0.05 to 0.08; 2 studies, 396 infants; low-certainty evidence). Only one study enrolling 79 infants reported components of the outcome moderate to severe neurodevelopmental disability at 18 to 26 months. The evidence is very uncertain about the effect of caffeine on cognitive developmental delay compared to other methylxanthines (RR 0.17, 95% CI 0.02 to 1.37; RD -0.12, 95% CI -0.24 to 0.01; 1 study, 79 infants; very low-certainty evidence). The evidence is very uncertain about the effect of caffeine on language developmental delay compared to other methylxanthines (RR 0.76, 95% CI 0.37 to 1.58; RD -0.07, 95% CI -0.27 to 0.12; 1 study, 79 infants; very low-certainty evidence). The evidence is very uncertain about the effect of caffeine on motor developmental delay compared to other methylxanthines (RR 0.50, 95% CI 0.13 to 1.96; RD -0.07, 95% CI -0.21 to 0.07; 1 study, 79 infants; very low-certainty evidence). The evidence is very uncertain about the effect of caffeine on visual and hearing impairment compared to other methylxanthines. At 24 months of age, visual impairment was seen in 8 out of 11 infants and 10 out of 11 infants in the caffeine and other methylxanthines groups, respectively. Hearing impairment was seen in 2 out of 5 infants and 1 out of 1 infant in the caffeine and other methylxanthines groups, respectively. No studies reported the outcomes cerebral palsy, gross motor disability, and mental development. Compared to other methylxanthines, caffeine may result in little to no difference in BPD/chronic lung disease, defined as 28 days of oxygen exposure at 36 weeks' postmenstrual age (RR 1.40, 95% CI 0.92 to 2.11; RD 0.04, 95% CI -0.01 to 0.09; 3 studies, 481 infants; low-certainty evidence). The evidence is very uncertain about the effect of caffeine on side effects (tachycardia, agitation, or feed intolerance) leading to a reduction in dose or withholding of methylxanthines compared to other methylxanthines (RR 0.17, 95% CI 0.02 to 1.32; RD -0.29, 95% CI -0.57 to -0.02; 1 study, 30 infants; very low-certainty evidence). Caffeine may result in little to no difference in duration of hospital stay compared to other methylxanthines (median (interquartile range): caffeine 43 days (27.5 to 61.5); other methylxanthines 39 days (28 to 55)).

No studies reported the outcome seizures. Authors' conclusions: Although caffeine has been shown to improve important clinical outcomes, in the few studies that compared caffeine to other methylxanthines, there might be little to no difference in mortality, bronchopulmonary dysplasia, and duration of hospital stay. The evidence is very uncertain about the effect of caffeine compared to other methylxanthines on long-term development and side effects. Although caffeine or other methylxanthines are widely used in preterm infants, there is little direct evidence to support the choice of which methylxanthine to use. More research is needed, especially on extremely preterm infants born before 28 weeks of gestation. Data from four ongoing studies might provide more evidence on the effects of caffeine or other methylxanthines.

PMID: 37791592

22. Magnetic resonance imaging sequences for children with spastic cerebral palsy: Is two better than one?

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No abstract available

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23. Positive and negative cell therapy in randomized control trials for central nervous system diseases

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Neurorestorative cell therapies have been tested to treat patients with nervous system diseases for over 20 years. Now it is still hard to answer which kinds of cells can really play a role on improving these patients' quality of life. Non-randomized clinical trials or studies could not provide strong evidences in answering this critical question. In this review, we summarized randomized clinical trials of cell therapies for central nervous diseases, such as stroke, spinal cord injury, cerebral palsy (CP), Parkinson's disease (PD), multiple sclerosis (MS), brain trauma, amyotrophic lateral sclerosis (ALS), etc. Most kinds of cell therapies demonstrated negative results for stoke, brain trauma and amyotrophic lateral sclerosis. A few kinds of cell therapies showed neurorestorative effects in this level of evidence-based medicine, such as olfactory ensheating cells for chronic ischemic stroke. Some kinds of cells showed positive or negative effects from different teams in the same or different diseases. We analyzed the possible failed reasons of negative results and the cellular bio-propriety basis of positive results. Based on therapeutic results of randomized control trials and reasonable analysis, we recommend: (1) to further conduct trials for successful cell therapies with positive results to increase neurorestorative effects; (2) to avoid in repeating failed cell therapies with negative results in same diseases because it is nonsense for them to be done with similar treatment methods, such as cell dosage, transplanting way, time of window, etc. Furthermore, we strongly suggest not to do non-randomized clinical trials for cells that had shown negative results in randomized clinical trials.

PMID: 37783557

24. Association of epilepsy with neuroimaging patterns in children with cerebral palsy

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Objectives: In this study, we examined whether epilepsy and drug-resistant epilepsy are associated with neuroimaging findings in children with cerebral palsy (CP). Methods: Magnetic resonance imaging classification system (MRICS) proposed by Surveillance of Cerebral Palsy in Europe (SCPE) was used for classification of different MRI patterns in patients with cerebral palsy. We reviewed the brain MRI scans and medical records of children with CP who were followed-up in our clinic between 2019 and 2023. Patients were divided into three categories: CP without epilepsy, CP with controlled epilepsy and CP with DRE. MRI patterns were grouped as maldevelopments, predominant white matter injury, predominant gray matter injury, miscellaneous (delayed myelination, cerebral atrophy, cerebellar atrophy, brainstem lesions and calcifications, lesions that were not classified under any other group) and normal according to MRICS of the SCPE. Results: There were 325 CP patients. The most common MRI patterns were predominant white matter injury (47.6%) and gray matter injury (23.8%). There was a 1.5-fold reduction in the risk of epilepsy in patients with predominant white matter injury (OR = 1.54, 95% CI 1.23-1.94). In contrast, children in the miscellaneous group had significantly higher risks of epilepsy ($p < 0.001$), and we were able to determine that miscellaneous findings increased the risk by 1.8 times (OR = 1.77, 95% CI 1.47-2.12). Conclusion: In conclusion, more than half of the children with CP had epilepsy, 40.7% of whom had DRE. On MRI, miscellaneous findings may indicate a poor prognosis for epilepsy, while predominant white matter injury may indicate a good outcome. Children with CP, especially those with miscellaneous findings on MRI, should be closely monitored for epilepsy development.

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