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Cerebral palsy research news

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Interventions and Management

1. The Efficacy of Two Models of Intensive Upper Limb Training on Health-Related Quality of Life in Children with Hemiplegic Cerebral Palsy Mainstreamed in Regular Schools: A Double-Blinded, Randomized Controlled Trial
Hasan Bingol, Mintaze Kerem Gunel, Halil Alkan

Physiother Theory Pract. 2021 Nov 7;1-16. doi: 10.1080/09593985.2021.1999355. Online ahead of print.

Background: Children with hemiplegic cerebral palsy (CP) are at risk of reduced upper limb function and poorer quality of life than their typically developing peers. Although upper limb impairments have potential negative impact on the health-related quality of life (HRQOL) in children with hemiplegic CP, the efficacy of upper limb rehabilitation approaches aiming to improve upper limb impairments on HRQOL has not been adequately investigated. Objective: This study compares the efficacy of two modes of activity-based upper limb rehabilitation (modified constraint-induced movement therapy [mCIMT] and bimanual training [BIM]) on HRQOL outcomes in children with hemiplegic CP mainstreamed in regular schools. Methods: Thirty children with hemiplegic CP aged between 7 and 11 years (mean age 8.53 ± 1.54 years) who had functional ability levels of I-III according to the Manual Ability Classification System (MACS), Gross Motor Classification System (GMFCS), and Communication Function Classification System (CFCS) were randomly assigned to receive mCIMT or BIM training. Result: Both mCIMT and BIM yielded statistically significant improvements in all HRQOL domains immediately after the 10-week interventions ($P < .001$), which were retained at 16 weeks. Conclusion: While there were some differences in the intervention effects, both of these upper limb rehabilitation approaches based on intensive unimanual or bimanual activity may be beneficial for improving perceived life satisfaction related to physical activity, general mood, family, friends, and school among children with hemiplegic CP mainstreamed in regular schools.

PMID: 34743663

2. Somatosensory Plasticity in Hemiplegic Cerebral Palsy Following Constraint Induced Movement Therapy
Cecilia Jobst, Samantha J D'Souza, Natasha Causton, Sabah Master, Lauren Switzer, Douglas Cheyne, Darcy Fehlings

Pediatr Neurol. 2021 Sep 30;126:80-88. doi: 10.1016/j.pediatrneurol.2021.09.019. Online ahead of print.

Background: Children with hemiplegic cerebral palsy (HCP) experience upper limb somatosensory and motor deficits. Although constraint-induced movement therapy (CIMT) improves motor function, its impact on somatosensory function remains underinvestigated. Objective: The objective of this study was to evaluate somatosensory perception and related brain responses in children with HCP, before and after a somatosensory enhanced CIMT protocol, as measured using clinical sensory and motor assessments and magnetoencephalography. Methods: Children with HCP attended a somatosensory enhanced CIMT camp. Clinical somatosensory (tactile registration, 2-point discrimination, stereognosis, proprioception, kinesthesia) and motor outcomes (Quality of Upper Extremity Skills [QUEST] Total/Grasp, Jebsen-Taylor Hand Function

Test, grip strength, Assisting Hand Assessment), as well as latency and amplitude of magnetoencephalography somatosensory evoked fields (SEF), were assessed before and after the CIMT camp with paired sample t-tests or Wilcoxon signed-rank tests. Results: Twelve children with HCP (mean age: 7.5 years, standard deviation: 2.4) participated. Significant improvements in tactile registration for the affected (hemiplegic) hand (Z = 2.39, P = 0.02) were observed in addition to statistically and clinically significant improvements in QUEST total (t = 3.24, P = 0.007), QUEST grasp (t = 3.24, P = 0.007), Assisting Hand Assessment (Z = 2.25, P = 0.03), and Jebsen-Taylor Hand Function Test (t = -2.62, P = 0.03). A significant increase in the SEF peak amplitude was also found in the affected hand 100 ms after stimulus onset (t = -2.22, P = 0.04). Conclusions: Improvements in somatosensory clinical function and neural processing in the affected primary somatosensory cortex in children with HCP were observed after a somatosensory enhanced CIMT program. Further investigation is warranted to continue to evaluate the effectiveness of a sensory enhanced CIMT program in larger samples and controlled study designs.

PMID: 34742103

3. Shortening wrist arthrodesis using a Rush pin in adult spastic wrist: a series of 15 cases
Victor Lestienne, Valentin Oca, Alexandre Cornu, Christian Fontaine, Etienne Allart, Nadine Sturbois-Nachef

Hand Surg Rehabil. 2021 Nov 6;S2468-1229(21)00622-8. doi: 10.1016/j.hansur.2021.09.011. Online ahead of print.

In spastic patients, shortening wrist arthrodesis (SWA) is indicated in cases of severe fixed flexion contracture. At present, the most commonly used technique is dorsal plate osteosynthesis. Ideally, fixation with smaller hardware volume farther from the tendons would limit postoperative tendon irritation and reoperation rates for hardware removal. The objective of our study was to evaluate the efficacy of Rush pin SWA in adults. A retrospective study included all patients with a central neurological impairment, undergoing SWA using a Rush pin inserted through the head of the third metacarpal, and with at least 6 months' follow-up. Attainment of preoperative objectives was evaluated by Global Assessment of Response to Treatment (GART, ranging from -4 to +4) and, for functional objectives, the House score and the Frenchay Arm Test. Consolidation and any degenerative changes in the third metacarpophalangeal joint were assessed on X-ray. Fifteen patients were included, with a mean follow-up of 13 months (range, 6-29). In general, the preoperative objectives were attained: mean GART score was 2.7 (range, 1-4). Functional objectives were attained in 3 of the 11 patients followed up (27%). In all cases, the arthrodesis had healed at a mean 74 days (range, 39-102). Three had hardware removed after consolidation; 1 experienced discomfort at the head of the third metacarpal. Rush pin arthrodesis is an interesting alternative to plate arthrodesis in the management of severe wrist flexion contracture in spastic patients. It gives satisfactory results with regard to preoperative objectives and is not associated with complications. LEVEL OF EVIDENCE: IV, retrospective study without control group.

PMID: 34752970

4. Rhythm and Reaching: The Influence of Rhythmic Auditory Cueing in a Goal-Directed Reaching Task With Adults Diagnosed With Cerebral Palsy
Jacqueline C Ladwig, Tamires C do Prado, Stephanie J Tomy, Jonathan J Marotta, Cheryl M Glazebrook

Adapt Phys Activ Q. 2021 Nov 5;1-16. doi: 10.1123/apaq.2021-0019. Online ahead of print.

Improvements in functional reaching directly support improvements in independence. The addition of auditory inputs (e.g., music, rhythmic counting) may improve goal-directed reaching for individuals with cerebral palsy (CP). To effectively integrate auditory stimuli into adapted teaching and rehabilitation protocols, it is necessary to understand how auditory stimuli may enhance limb control. This study considered the influence of auditory stimuli during the planning or execution phases of goal-directed reaches. Adults (with CP = 10, without CP = 10) reached from a home switch to two targets. Three conditions were presented-no sound, sound before, and sound during-and three-dimensional movement trajectories were recorded. Reaction times were shorter for both groups in the sound before condition, while the group with CP also reached peak velocity relatively earlier in the sound before condition. The group with CP executed more consistent movements in both sound conditions. Sound presented before movement initiation improved both the planning and execution of reaching movements for adults with CP.

PMID: 34740992

5. Long-term effects of spasticity treatment, including selective dorsal rhizotomy, for individuals with cerebral palsy
Bruce A MacWilliams, Mark L McMulkin, Elizabeth A Duffy, Meghan E Munger, Brian Po-Jung Chen, Tom F Novacheck, Michael H Schwartz, Selective Dorsal Rhizotomy Outcomes Research Team

Dev Med Child Neurol. 2021 Nov 10. doi: 10.1111/dmcn.15075. Online ahead of print.

Aim: To understand the long-term effects of comprehensive spasticity treatment, including selective dorsal rhizotomy (SDR), on individuals with spastic cerebral palsy. Method: This was a pre-registered, multicenter, retrospectively matched cohort study. Children were matched on age range and spasticity at baseline. Children at one center underwent spasticity treatment including SDR (Yes-SDR, n=35) and antispastic injections. Children at two other centers had no SDR (No-SDR, n=40 total) and limited antispastic injections. All underwent subsequent orthopedic treatment. Participants returned for comprehensive long-term assessment (age ≥21y, follow-up ≥10y). Assessment included spasticity, contracture, bony alignment, strength, gait, walking energy, function, pain, stiffness, participation, and quality of life. Results: Spasticity was effectively reduced at long-term assessment in the Yes-SDR group and was unchanged in the No-SDR group. There were no meaningful differences between the groups in any measure except the Gait Deviation Index (Yes-SDR + 11 vs No-SDR + 5) and walking speed (Yes-SDR unchanged, No-SDR declined 25%). The Yes-SDR group underwent more subsequent orthopedic surgery (11.9 vs 9.7 per individual) and antispastic injections to the lower limbs (14.4 vs <3, by design). Interpretation: Untreated spasticity does not cause meaningful impairments in young adulthood at the level of pathophysiology, function, or quality of life.

PMID: 34755903

6. Prepontine placement of an intrathecal baclofen pump catheter for treatment of dystonia
Thomas Gianaris, Ryan M Holland, Nicolas W Villelli, Albert E Lee

Case Reports Surg Neurol Int. 2021 Sep 30;12:477. doi: 10.25259/SNI_146_2019. eCollection 2021.

Background: Cerebral palsy with medically refractory spasticity and dystonia is a condition that often benefits from intrathecal baclofen pump therapy to treat these symptoms. In this case report, an intracranial baclofen catheter was placed in the prepontine space to improve withdrawal symptoms in a patient unable to undergo new lumbar catheter placement due to infection. Case description: A 22-year-old female with past medical history of cerebral palsy presented with baclofen pump failure and was unable to undergo placement of a new lumbar baclofen catheter due to an infection in her lower back precluding safe and efficacious catheter placement. It was decided the patient would benefit from intrathecal baclofen administered in the prepontine space as a means to avoid a lumbar catheter and thus bypass this prior infection site. An endoscopic third ventriculostomy (ETV) was performed with the endoscope and the distal end of the baclofen pump catheter was fed through this ETV into the prepontine space. Placement in the prepontine space was confirmed by a follow-up head computed tomography. There was a significant improvement in autonomic symptoms and spasticity. By postoperative day 5, the patient was surgically and medically cleared for discharge. Conclusion: In cases of severe baclofen withdrawal due to dysfunctional pumps, immediate reversal is preferred but may not be feasible due to factors such as infection. This case report has demonstrated that prepontine catheter placement can be effective for the administration of baclofen to reverse withdrawal symptoms in these types of patients.

PMID: 34754527

7. The Foot in Cerebral Palsy
Julieanne P Sees, Freeman Miller

Review Foot Ankle Clin. 2021 Dec;26(4):639-653. doi: 10.1016/j.fcl.2021.07.002. Epub 2021 Aug 17.

Children with cerebral palsy frequently develop foot deformities, most commonly equinus contractures, which can be managed with orthotics up to age 5 to 7 years. Plantar flexor lengthening, typically around this age, should be restricted to the offending muscle only, usually with a fascia release of the gastrocnemius. Equinovarus, mainly a problem in children with unilateral cerebral palsy, often responds to plantar flexor lengthening. If further tendon transfers are needed, they should be done when the child is older to avoid overcorrection. Planovalgus mostly improves spontaneously up to age 5 years. Surgical correction is best done in adolescence.

PMID: 34752232

8. Effects of Power Training on Gait, Power, and Function in Children with Cerebral Palsy
Michaella Drumm, Julia Fabiano, Evelyn Lee, Jennifer Jezequel, Ashwini K Rao, Lisa Yoon

Phys Occup Ther Pediatr. 2021 Nov 9;1-15. doi: 10.1080/01942638.2021.1995098. Online ahead of print.

Aims: This scoping review aims to: 1) examine available literature regarding the effects of power training on gait speed, power, and function in ambulatory children with CP and 2) identify the variations in exercise dosage and rehabilitation recommendations for power training and plyometrics in children with CP. Methods: Four databases (PubMed, CINAHL, Embase, and Cochrane) were searched for papers including power or plyometric training with outcome measures for gait, power or functional performance. ES was calculated for RCTs. Cohorts and case series/studies were evaluated qualitatively. Results: Ten articles fit search criteria: four RCTs, three cohort studies, one case series, and two case studies. Power training consistently demonstrated improvements in muscle power compared to its effects on gait and function. ES of mean MPST (W) ranged from 0.36-1.13. 1 MWT and SSGS ES were 1.31 and 1.15, respectively. TUG ES ranged from -0.33 to -2.42. ES for GMFM-66 was 0.13 and 1.11 for Dimension D and Dimension E, respectively. Conclusions: There is limited, but promising evidence to support that power training may improve gait speed, power, and function in children with CP. Future, more robust research is required to examine effects in a larger, diverse population, to determine long-term effects and exercise prescription.

PMID: 34753380

9. AllPlay Dance: Two Pilot Dance Projects for Children With Disability, Developed and Assessed With a Dance Studies Approach
Olivia Millard, Ebony Lindor, Nicole Papadopoulos, Carmel Sivaratnam, Jane McGillivray, Nicole Rinehart

Front Psychol. 2021 Oct 22;12:567055. doi: 10.3389/fpsyg.2021.567055. eCollection 2021.

AllPlay Dance is founded on a collaborative approach to research between the School of Psychology and the School of Communication of Creative Arts, both of Deakin University. The research is also undertaken in partnership with professional ballet company, Queensland Ballet. This paper describes the development and execution of two pilot projects for children with disability, utilizing a dance studies methodology. The projects were conducted in 2018 and 2019 for children with cerebral palsy (CP) and autism spectrum disorder, as part of the AllPlay Dance program. Participants with disabilities ranged in age from 7 to 12 years. As well as describing the approach to the program development, we discuss the involvement of older and more experienced buddies who were included as a method to support the participation in dance of children with disabilities. We will also describe the diffusion of authorship in the making of group dances as a tool for inclusion and the premise of dance as a social practice in which participants inter-subjectively generate meaning and sense making. The AllPlay Dance projects were developed as a series of dance classes in which participants worked with set or learned movement material, dance improvisation, and tasks for movement generation in order to collectively generate a dance for performance. This paper focuses on the aim of developing inclusive approaches to dance classes that challenge "ableist" notions of dance as spectacle to enable to work toward building transferable programs to allow all children who so desire and to participate in dance in their communities.

PMID: 34751219

10. Effects of Piano Training in Unilateral Cerebral Palsy Using Probabilistic and Deterministic Tractography: A Case Report
Ana Alves-Pinto, Mónica Emch, Renée Lampe

Case Reports Front Hum Neurosci. 2021 Oct 21;15:622082. doi: 10.3389/fnhum.2021.622082. eCollection 2021.

Cerebral palsy (CP) is an umbrella term encompassing motor and often additional disabilities, resulting from insult to the developing brain and remaining throughout life. Imaging-detected alterations in white matter microstructure affect not only motor but also sensorimotor pathways. In this context, piano training is believed to promote sensorimotor rehabilitation for the multiplicity of skills and neuronal processes it involves and integrates. However, it remains unknown how this contribution may occur. Here, effects of 1.5 years of piano training in an adolescent with unilateral CP were investigated through tests of manual function and by comparing fractional anisotropy, mean diffusivity, radial and axial diffusivity in neuronal pathways pre

- vs. post-training. In the absence of a control condition and of data from a larger cohort, both probabilistic neighborhood and deterministic tractography were employed to reduce bias associated with a single-case analysis and/or with user-input. No changes in manual function were detected with the tests performed. In turn, the two tractography methods yielded similar values for all studied metrics. Furthermore, post-hoc analyses yielded increased fractional anisotropy accompanied by decreases in mean diffusivity in the bilateral dorsal cingulate that were at least as large as and more consistent than in the bilateral corticospinal tract. This suggests contributions of training to the development of non-motor processes. Reduced anisotropy and correspondingly high mean diffusivity were observed for the bilateral corticospinal tract as well as for the right arcuate and the inferior longitudinal fasciculus, two sensory processing-related pathways, confirming the importance of sensorimotor rehabilitation in CP.

PMID: 34744658

11. Phenotype assessment in neurologically impaired paediatric patients: Impact of a nutrition intervention protocol
Vasiliki Katseni, Euthymia Vargiami, Thomais Karagiozoglou-Lampoudi, Efstratia Daskalou, Dimitrios Zafeiriou

Clin Nutr. 2021 Oct 23;40(12):5734-5741. doi: 10.1016/j.clnu.2021.10.011. Online ahead of print.

Background: Deficits in nutritional status and functional feeding disorders are common in Neurologically Impaired Paediatric Patients (NIPP). Interventions addressing these problems could offer better overall health status and quality of life in this group of patients, but the extent of their effectiveness is yet to be determined. Recent guidelines concerning the nutritional care of NIPP have been published from ESPGHAN but compliance to them has not been assessed. Aim: The study aimed to assess the phenotypic profile of a group of NIPP attending the outpatient clinic of a pediatric department, and to implement, for the first time to our knowledge, an individualized nutritional intervention protocol following ESPGHAN guidelines 2017 as well as to assess the impact on phenotypic parameters and nutritional status. Patients and methods: 68NIPP and their caregivers aged 1m-17 years (83.8% suffering from cerebral palsy (CP) were invited to assess their phenotypic parameters and to implement in a nutrition intervention protocol in order to improve their dietary intake and nutritional status. Anthropometry (weight, height, triceps skinfold thickness, mid upper arm circumference) was expressed as z-scores for age and sex using WHO Anthro software and classified following the WHO criteria. Gross Motor Function Classification System (GMFCS), Manual Ability Classification System (MACS), Dysphagia Disorder Survey (DDS), Saliva Severity Scale (SSS), gastrointestinal complications, energy and nutrient intake were assessed at the beginning (zero point), after 6 (point 1) and 12 (point 2) months period. Intake to Requirement ratio (I/R) was derived. At zero point, following the baseline evaluation, caregivers were advised and educated on nutrition protocol and customized nutrition plans were handed out. The impact of the nutritional intervention on the phenotypic parameters was recorded on follow up visits (points 1, 2).The primary outcomes analyzed were anthropometric parameters (Waz), as indicators of nutritional status. GMFCS, MACS, DDS, SSS, FA were evaluated as possible predictors of this outcome. Secondary outcomes included the impact of the intervention protocol on the phenotypic parameters during the study period. Results: Based on weight for age z-score (Waz ≤ -2) (WHO) 17 patients (32.1%) were undernourished, 5/68 (10, 4%) were with triceps skinfold thickness z-score (TSTz) <-2 and 3/68 (7%) with mid upper arm circumference z-score (MUACz) <-2. Z-scores (WHO) for weight (p1 = 0, 036) (p2 = 0, 003), body mass index (BMI) (p2 = 0,000), MUAC (p1 = 0, 029) and TST (p1 = 0, 021) (p3 = 0, 044) were significantly improved in follow-up evaluations compared to the baseline. Less NIPP were found to be underweight according to Waz from point 1 to point 2 (p3 = 0, 006), as well as stunding according to height for age z-score (Haz) from point 1 to point 2 (p ≤ 0,001). Patients with higher levels of GMFCS (p1 = 0,040), MACS (p1 = 0,028) DDS (p1 = 0,001) and SSS (p1 = 0,005) had significantly lower Haz. Patients with higher levels of SSS (p1 = 0,002) had significantly lower TSTz scores. There were no significant changes in the classification of NIPP according to DDS or the patients' feeding ability. The energy (kcals) intake/kg of body weight (bw) was significantly higher at point 2 compared to point zero (p3 = 0,028), protein intake/kg of body weight was significantly higher at points 1 and 2 compared to point zero (p1 = 0,026, p3 = 0,003), and fat intake/kg of body weight (bw) was significantly higher at point 2 compared to point zero (p3 = 0,012). Intake of energy (kcals)/bw (p1 = 0,026), (p2 = 0,046), (p3 = 0,048) carbs/bw (p1 = 0,014) (p2 = 0,042), I/R of pro (p1 = 0,032), (p3 = 0,013), and fat/kg (p2 = 0, 033) (p3 = 0,037) were found to be significantly lower in higher GMFCS levels. DQI did not improve during the study period nor correlated to any of the anthropometric parameters. Gastrointestinal complications correlated with Waz (r = -, 285 p1 = 0, 011). Feeding Ability (FA) was found to be the only strong predictor for Waz at baseline evaluation (p = 0,012) when a multiple regression was run along with DDS. Conclusion: Underweight was detected in one third of the patients, some degree of dysphagia in 69% and gastrointestinal complications in 58.8% of the sample. Height for age z-score (Haz) was the anthropometric parameter most sensitive to the changes in ranking on motor and functional feeding scores. The implementation of a customized nutrition intervention protocol in line with ESPGHAN's guidelines had a beneficial effect on improving dietary intake and nutritional status of NIPP after a 12 months period. Better results could be expected if dysphagia and feeding ability were also addressed by appropriate intervention protocols. Patients' feeding ability is of importance for predicting Waz.

PMID: 34753089

12. Defining Persistent Total Parenteral Nutrition Use in Patients with Neurologic Impairment
Taylor Jersak, Stephani S Kim, Garey Noritz, Marissa Testa, Lisa Humphrey

J Palliat Med. 2021 Nov 10. doi: 10.1089/jpm.2021.0086. Online ahead of print.

Background: Patients with neurologic impairment (NI) experience gastrointestinal symptoms as one of three common problems associated with NI, including occasional persistent total parenteral nutrition (TPN) use. Objective: To describe the incidence of persistent TPN use in patients with NI. Design: Retrospective chart review on patients 0-38 years old enrolled in the Complex Health Care Program from January 2011 to October 2015. Setting/Subjects: This study occurred in a United States pediatric tertiary care hospital. Two hundred and eight participants were included based on NI, utilizing a surgical feeding tube, and having encounters with a dietitian. Measurements: The primary outcome was incidence of persistent TPN use in patients with NI. Secondary outcomes included mortality rate, hospitalization frequency, time-to-TPN initiation, and describing symptoms preceding persistent TPN use. Results: Median number of admissions was 4 for 168 hospitalized patients (59% male, 58% White). One hundred twenty-five patients required admission for unplanned bowel rest with average length-of-stay of 7.3 days. Twenty-six patients required TPN initiation. Average time-to-TPN was two years since enrollment. Mortality rate was 14% (n = 28). TPN initiation (odds ratio [OR]: 3.99; 95% confidence interval [CI]: 1.16-13.8) was significantly associated with increased OR of mortality. Conclusions: Our study demonstrates a substantial population of patients with NI and surgical feeding tube are affected by persistent feeding intolerance. We propose that persistent TPN use may be a risk factor for mortality. Additional research is needed to delineate relationships between persistent TPN use, hospitalizations, and mortality.

PMID: 34757811

13. Satisfaction of Oral Health Education among Parents and Caregivers Regarding Children with Special Healthcare Needs in Riyadh, Saudi Arabia
Altaf H Shah, Mohammad Al Refeai, Faisal M Alolaywi, Shabnam Gulzar, Faris Y Asiri, Al Bandary Al Jameel

J Contemp Dent Pract. 2021 Aug 1;22(8):894-899.

Aim and objective: The aim of the study was to evaluate the satisfaction of an oral health promotion program among parents and caregivers of children with special healthcare needs (CSHCN) in Riyadh, Saudi Arabia. Materials and methods: A digital survey using Google forms was used during an event to commemorate the World Disability Day. Stalls for oral health education and training were set up at Disabled Children Society, Riyadh, Saudi Arabia. Oral health education was carried out using written pamphlets, brochures, and videos. Live demonstration on dental models was used for tooth-brushing training using a powered toothbrush. Parents and caregivers were asked to complete a survey about oral health education during the event. An overall 189 parents and caregivers of CSHCN including 62 males and 127 females completed the survey. The responses were entered digitally prior to being evaluated. Results: The respondents were parents and/or caregivers of children mostly having cerebral palsy followed by autism. Females consisted of 67% of the respondents. Eight-one percent of respondents rated it as highly satisfied. Ninety-one percent of the respondents perceived that the information provided was new for them regarding oral healthcare for CSHCN. Ninety-eight percent of respondents were likely to attend a similar event in the future. Conclusion: Majority of parents and caregivers of CSHCN were highly satisfied by the oral health education during the event. They felt that they can take better care of the oral health of their CSHCN after the oral health education and training. Clinical significance: Oral health among CSHCN is neglected when compared to children among the general population. CSHCN exhibit many barriers to oral health care. Oral health promotion among CSHCN is important as they have a high unmet oral health need.

PMID: 34753841

14. Continuing decline in the prevalence of cerebral palsy in Denmark for birth years 2008-2013
Mads Langager Larsen, Gija Rackauskaite, Gorm Greisen, Bjarne Laursen, Peter Uldall, Lone Krebs, Christina Engel Hoei-Hansen

Eur J Paediatr Neurol. 2020 Oct 17;S1090-3798(20)30196-3. doi: 10.1016/j.ejpn.2020.10.003. Online ahead of print.

Aim: To quantify and analyse the prevalence and clinical features of cerebral palsy (CP) in Denmark for birth years 2008-2013 and compare results with previous periods. Method: A nationwide register-based study covering all children with a confirmed diagnosis of CP born in Denmark. Information about CP subtype, aetiology and severity was collected from the Cerebral Palsy Follow-up Program and supplemented from medical files. Data from the Danish Medical Birth Register was included, and the results were compared to previous data from the Danish National Cerebral Palsy Register. Prevalence per 1000 live births and proportions were analysed using the Cochran-Armitage test for trend. Results: The period covered 368,618 live births and 636 children with CP, making the overall prevalence for the period 1.73 per 1000 live births. This was significantly lower than the prevalence of 1.99 for the previous period 1999-2007 ($p = 0.004$). The decline in prevalence between the two periods was mainly due to a decrease in children with bilateral spastic and dyskinetic CP born after 37 gestational weeks. The decline in prevalence was accompanied by a smaller proportion of children with associated impairment. Conclusion: We found a decrease in prevalence and severity in CP among Danish children. The decline was most pronounced in children born after 37 gestational weeks with severe subtypes of CP. National guidelines that recommend induction of labour before the completion of week 42 and therapeutic hypothermia for term neonates with hypoxic-ischaemic encephalopathy, may have contributed to the decline.

PMID: 34756357

15. Functional Classification of Children with Cerebral Palsy in Krapina-Zagorje Country
Sunčica Martinec, Gordana Cesarec, Ana Marija Tomečak Krilić, Tomislav Radošević, Žarko Bakran, Vlatka Mejaški Bošnjak

Acta Clin Croat. 2021 Jun;60(2):282-289. doi: 10.20471/acc.2021.60.02.15.

The aim was to study functional abilities and to create functional classification of children with cerebral palsy (CP) in Krapina-Zagorje County, based on the classification of gross and fine motor skills and associated impairments. Classification was performed according to the SCPE (Surveillance of Cerebral Palsy in Europe) criteria. We used standardized and complementary functional classification systems for cerebral palsy to create a functional profile. Research included 44 children with CP in the age range of 4 to 18 years. The results showed that the majority of children had bilateral spastic CP (63.6%), followed by unilateral spastic (22.7%) while the representation of dyskinetic CP was 9.09% and ataxic CP 4.55%. Based on the classification of gross and fine motor skills, 43.2% of children had the ability to walk, 11% of children could walk with assistive mobility devices, while 45.4% of children had a low functional level. The study also analyzed the associated impairments where higher classification score of motor impairment correlated with the severity of impairment. The results showed that children with dyskinetic CP and severe motor impairment could have mild cognitive impairment. We systematically present the neuropsychological and functional profile according to the CP type.

PMID: 34744279

16. Living Conditions and Social Outcomes in Adults With Cerebral Palsy
Katina Pettersson, Elisabet Rodby-Bousquet

Front Neurol. 2021 Oct 21;12:749389. doi: 10.3389/fneur.2021.749389. eCollection 2021.

Objectives: To analyse the living conditions and social outcomes (housing, engagement in employment or higher education, access to personal assistance and having a partner) in adults with cerebral palsy (CP) relative to their age, sex, communication ability, and motor skills. Methods: Cross-sectional registry-based study of 1,888 adults (1,030 males/858 females) with CP in the Swedish CP follow-up programme, median age 25 years (range 16-78 y). Type of housing, occupation, access to personal assistance and having a partner were analysed relative to their age, sex, and the classification systems for Gross Motor Function (GMFCS) and Communication Function (CFCS). Binary logistic regression models were used to calculate odds ratios (OR) for independent living, competitive employment, and having a partner. Results: Most of the 25- to 29-year olds (55.6%) lived independently, increasing to 72.4% in 40- to 49-year olds, while the majority (91.3%) of those under 20 years lived with their parents. Independent living was almost equal in adults at GMFCS levels I (40.2%) and V (38.6%). This parity was explained by access to personal assistance, which increased with higher GMFCS and CFCS levels. Personal assistance of >160 hours/week was associated with a high probability of independent living (OR 57). In the age span 20-64 years, 17.5% had competitive employment and 45.2% attended activity centres for people with intellectual disabilities. In the younger age group up to 24 years old, 36.9% went to mainstream/higher education and 20.5% went to special schools. In total, 13.4% had a partner and 7.8% lived together. Slightly more women than men had a partner, and most individuals were classified at CFCS level I. Conclusion: Only one in eight adults with CP has a partner, and one in six has competitive employment. Access to

personal assistance is the single most important factor for independent living. It is vital to support adults with CP throughout their lifespan to achieve the best possible outcomes in all aspects of life.

PMID: 34744986

17. Late (≥ 7 days) systemic postnatal corticosteroids for prevention of bronchopulmonary dysplasia in preterm infants
Lex W Doyle, Jeanie L Cheong, Susanne Hay, Brett J Manley, Henry L Halliday

Review Cochrane Database Syst Rev. 2021 Nov 11;11:CD001145. doi: 10.1002/14651858.CD001145.pub5.

Background: Many infants born preterm develop bronchopulmonary dysplasia (BPD), with lung inflammation playing a role. Corticosteroids have powerful anti-inflammatory effects and have been used to treat individuals with established BPD. However, it is unclear whether any beneficial effects outweigh the adverse effects of these drugs. Objectives: To examine the relative benefits and adverse effects of late (starting at seven or more days after birth) systemic postnatal corticosteroid treatment for preterm infants with evolving or established BPD. Search methods: We ran an updated search on 25 September 2020 of the following databases: CENTRAL via CRS Web and MEDLINE via OVID. We also searched clinical trials databases and reference lists of retrieved articles for randomised controlled trials (RCTs). We did not include quasi-RCTs. Selection criteria: We selected for inclusion in this review RCTs comparing systemic (intravenous or oral) postnatal corticosteroid treatment versus placebo or no treatment started at seven or more days after birth for preterm infants with evolving or established BPD. We did not include trials of inhaled corticosteroids. Data collection and analysis: We used standard Cochrane methods. We extracted and analysed data regarding clinical outcomes that included mortality, BPD, and cerebral palsy. We used the GRADE approach to assess the certainty of evidence. Main results: Use of the GRADE approach revealed that the certainty of evidence was high for most of the major outcomes considered, except for BPD at 36 weeks for all studies combined and for the dexamethasone subgroup, which were downgraded one level to moderate because of evidence of publication bias, and for the combined outcome of mortality or BPD at 36 weeks for all studies combined and for the dexamethasone subgroup, which were downgraded one level to moderate because of evidence of substantial heterogeneity. We included 23 RCTs (1817 infants); 21 RCTS (1382 infants) involved dexamethasone (one also included hydrocortisone) and two RCTs (435 infants) involved hydrocortisone only. The overall risk of bias of included studies was low; all were RCTs and most trials used rigorous methods. Late systemic corticosteroids overall reduce mortality to the latest reported age (risk ratio (RR) 0.81, 95% confidence interval (CI) 0.66 to 0.99; 21 studies, 1428 infants; high-certainty evidence). Within the subgroups by drug, neither dexamethasone (RR 0.85, 95% CI 0.66 to 1.11; 19 studies, 993 infants; high-certainty evidence) nor hydrocortisone (RR 0.74, 95% CI 0.54 to 1.02; 2 studies, 435 infants; high-certainty evidence) alone clearly reduce mortality to the latest reported age. We found little evidence for statistical heterogeneity between the dexamethasone and hydrocortisone subgroups (P = 0.51 for subgroup interaction). Late systemic corticosteroids overall probably reduce BPD at 36 weeks' postmenstrual age (PMA) (RR 0.89, 95% CI 0.80 to 0.99; 14 studies, 988 infants; moderate-certainty evidence). Dexamethasone probably reduces BPD at 36 weeks' PMA (RR 0.76, 95% CI 0.66 to 0.87; 12 studies, 553 infants; moderate-certainty evidence), but hydrocortisone does not (RR 1.10, 95% CI 0.92 to 1.31; 2 studies, 435 infants; high-certainty evidence) (P < 0.001 for subgroup interaction). Late systemic corticosteroids overall probably reduce the combined outcome of mortality or BPD at 36 weeks' PMA (RR 0.85, 95% CI 0.79 to 0.92; 14 studies, 988 infants; moderate-certainty evidence). Dexamethasone probably reduces the combined outcome of mortality or BPD at 36 weeks' PMA (RR 0.75, 95% CI 0.67 to 0.84; 12 studies, 553 infants; moderate-certainty evidence), but hydrocortisone does not (RR 0.98, 95% CI 0.88 to 1.09; 2 studies, 435 infants; high-certainty evidence) (P < 0.001 for subgroup interaction). Late systemic corticosteroids overall have little to no effect on cerebral palsy (RR 1.17, 95% CI 0.84 to 1.61; 17 studies, 1290 infants; high-certainty evidence). We found little evidence for statistical heterogeneity between the dexamethasone and hydrocortisone subgroups (P = 0.63 for subgroup interaction). Late systemic corticosteroids overall have little to no effect on the combined outcome of mortality or cerebral palsy (RR 0.90, 95% CI 0.76 to 1.06; 17 studies, 1290 infants; high-certainty evidence). We found little evidence for statistical heterogeneity between the dexamethasone and hydrocortisone subgroups (P = 0.42 for subgroup interaction). Studies had few participants who were not intubated at enrolment; hence, it is not possible to make any meaningful comments on the effectiveness of late corticosteroids in preventing BPD in non-intubated infants, including those who might in the present day be supported by non-invasive techniques such as nasal continuous positive airway pressure or high-flow nasal cannula oxygen/air mixture, but who might still be at high risk of later BPD. Results of two ongoing studies are awaited. Authors' conclusions: Late systemic postnatal corticosteroid treatment (started at seven days or more after birth) reduces the risks of mortality and BPD, and the combined outcome of mortality or BPD, without evidence of increased cerebral palsy. However, the methodological quality of studies determining long-term outcomes is limited, and no studies were powered to detect increased rates of important adverse long-term neurodevelopmental outcomes. This review supports the use of late systemic corticosteroids for infants who cannot be weaned from mechanical ventilation. The role of late systemic corticosteroids for infants who are not intubated is unclear and needs further investigation. Longer-term follow-up into late childhood is vital for assessment of important outcomes that cannot be assessed in early childhood, such as effects of late systemic corticosteroid treatment on higher-order neurological functions, including cognitive function, executive function, academic performance, behaviour, mental health, motor function, and lung function. Further RCTs of late systemic corticosteroids should include longer-term survival free of neurodevelopmental disability as the primary outcome.

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18. Association Between Brain Structural Network Efficiency at Term-Equivalent Age and Early Development of Cerebral Palsy in Very Preterm Infants
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Very preterm infants (born at less than 32 weeks gestational age) are at high risk for serious motor impairments, including cerebral palsy (CP). The brain network changes that antecede the early development of CP in infants are not well characterized, and a better understanding may suggest new strategies for risk-stratification at term, which could lead to earlier access to therapies. Graph theoretical methods applied to diffusion MRI-derived brain connectomes may help quantify the organization and information transfer capacity of the preterm brain with greater nuance than overt structural or regional microstructural changes. Our aim was to shed light on the pathophysiology of early CP development, before the occurrence of early intervention therapies and other environmental confounders, to help identify the best early biomarkers of CP risk in VPT infants. In a cohort of 345 very preterm infants, we extracted cortical morphometrics and brain volumes from structural MRI and also applied graph theoretical methods to diffusion MRI connectomes, both acquired at term-equivalent age. Metrics from graph network analysis, especially global efficiency, strength values of the major sensorimotor tracts, and local efficiency of the motor nodes and novel non-motor regions were strongly inversely related to early CP diagnosis. These measures remained significantly associated with CP after correction for common risk factors of motor development, suggesting that metrics of brain network efficiency at term may be sensitive biomarkers for early CP detection. We demonstrate for the first time that in VPT infants, early CP diagnosis is anteceded by decreased brain network segregation in numerous nodes, including motor regions commonly-associated with CP and also novel regions that may partially explain the high rate of cognitive impairments concomitant with CP diagnosis. These advanced MRI biomarkers may help identify the highest risk infants by term-equivalent age, facilitating earlier interventions that are informed by early pathophysiological changes.

PMID: 34758381

19. Health-Related Quality of Life among Children with Disabilities: Is There a Place for Parent-Proxy Reports? A Commentary on the "The Effects of Basic Photography Education on Quality of Life, Self-Esteem, Life Satisfaction and Moods in Children with Diplegic Cerebral Palsy: A Randomized Controlled Study"
Mariane Sentenac

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PMID: 34753386

20. Motor function and white matter connectivity in children cooled for neonatal encephalopathy
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Therapeutic hypothermia reduces the incidence of severe motor disability, such as cerebral palsy, following neonatal hypoxic-ischaemic encephalopathy. However, cooled children without cerebral palsy at school-age demonstrate motor deficits and altered white matter connectivity. In this study, we used diffusion-weighted imaging to investigate the relationship between white matter connectivity and motor performance, measured using the Movement Assessment Battery for Children-2, in children aged 6-8 years treated with therapeutic hypothermia for neonatal hypoxic-ischaemic encephalopathy at birth, who did not develop cerebral palsy (cases), and matched typically developing controls. Correlations between total motor scores and diffusion properties in major white matter tracts were assessed in 33 cases and 36 controls. In cases, significant correlations (FDR-corrected $P < 0.05$) were found in the anterior thalamic radiation bilaterally (left: $r = 0.513$; right: $r = 0.488$), the cingulate gyrus part of the left cingulum ($r = 0.588$), the hippocampal part of the left cingulum ($r = 0.541$), and the inferior fronto-occipital fasciculus bilaterally (left: $r = 0.445$; right: $r = 0.494$). No significant correlations were found in controls. We then constructed structural connectivity networks, for 22 cases and 32 controls, in which nodes represent brain regions and edges were determined by probabilistic tractography and weighted by fractional anisotropy. Analysis of whole-brain network metrics revealed correlations (FDR-corrected $P < 0.05$), in cases, between total motor scores and average node strength ($r = 0.571$), local efficiency ($r = 0.664$), global efficiency ($r = 0.677$), clustering coefficient ($r = 0.608$), and characteristic path

length (r = -0.652). No significant correlations were found in controls. We then investigated edge-level association with motor function using the network-based statistic. This revealed subnetworks which exhibited group differences in the association between motor outcome and edge weights, for total motor scores (P = 0.0109) as well as for balance (P = 0.0245) and manual dexterity (P = 0.0233) domain scores. All three of these subnetworks comprised numerous frontal lobe regions known to be associated with motor function, including the superior frontal gyrus and middle frontal gyrus. The subnetwork associated with total motor scores was highly left-lateralised. These findings demonstrate an association between impaired motor function and brain organisation in school-age children treated with therapeutic hypothermia for neonatal hypoxic-ischaemic encephalopathy.

PMID: 34749285