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Cerebral palsy research news

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Interventions and Management

1. Feasibility of At-Home Hand Arm Bimanual Intensive Training in Virtual Reality: Case Study

James E Gehringer, Anne Woodruff Jameson, Hailey Boyer, Jennifer Konieczny, Ryan Thomas, James Pierce Iii, Andrea B Cunha, Sandra Willett

Case Reports JMIR Form Res. 2024 Sep 6:8:e57588. doi: 10.2196/57588.

This single-participant case study examines the feasibility of using custom virtual reality (VR) gaming software in the home environment for low-dose Hand Arm Bimanual Intensive Training (HABIT). A 10-year-old with right unilateral cerebral palsy participated in this trial. Fine and gross motor skills as well as personal goals for motor outcomes were assessed before and after the intervention using the Box and Blocks Test, Nine-Hole Peg Test, and Canadian Occupational Performance Measure. Movement intensities collected via the VR hardware accelerometers, VR game scores, and task accuracy were recorded via the HABIT-VR software as indices of motor performance. The child and family were instructed to use the HABIT-VR games twice daily for 30 minutes over a 14-day period and asked to record when they used the system. The child used the system and completed the 14-hour, low-dose HABIT-VR intervention across 22 days. There was no change in Box and Blocks Test and Nine-Hole Peg Test scores before and after the intervention. Canadian Occupational Performance Measure scores increased but did not reach the clinically relevant threshold, due to high scores at baseline. Changes in motor task intensities during the use of VR and mastery of the VR bimanual tasks suggested improved motor efficiency. This case study provides preliminary evidence that HABIT-VR is useful for promoting adherence to HABIT activities and for the maintenance of upper extremity motor skills in the home setting.

PMID: 39241226

2. Pairing transcutaneous vagus nerve stimulation with an intensive bimanual training in children and adolescents with cerebral palsy: study protocol of a randomized sham-controlled trial

Viola Oldrati, Verusca Gasparroni, Arianna Michelutti, Andrea Ciricugno, Renato Borgatti, Simona Orcesi, Elisa Fazzi, Alessandra Morandi, Jessica Galli, Luigi Piccinini, Cristina Maghini, Maria Arioli, Zaira Cattaneo, Cosimo Urgesi, Alessandra Finisguerra

Front Neurol. 2024 Aug 16:15:1441128. doi: 10.3389/fneur.2024.1441128. eCollection 2024.

Background: Gross motor function impairments and manual dexterity deficits are frequently observed in children and adolescents with Cerebral Palsy (CP), having a major impact on their activity level and autonomy. Improving manual dexterity and activity level of patients with CP is often the focus of rehabilitation. Novel and adjuvant treatment methods that could support the standard training also in chronic conditions are a research priority. The transcutaneous Vagus Nerve Stimulation (tVNS) is a non-invasive brain stimulation technique, which provides a bottom-up stimulation of subcortical and cortical brain structures, enhancing brain GABA and Noradrenaline levels. This technique may play a pivotal role in brain plasticity, which has not been tested in CP patients before. Methods: 44 children and adolescents with CP will be involved, treated in pairs in a randomized, double-blind, pre-post test study. The two groups will undergo the Hand-Arm Bimanual Intensive Therapy Including Lower Extremities (HABIT-ILE) for 2 consecutive weeks, with 3 h daily sessions for 5 days per

week, for an overall time interval of 30 h; the training will be combined with the application for 75 min/day of active or sham tVNS, in separate, randomly allocated groups. The primary outcome measure will include the scores at the Assisting Hand Assessment and Box and Block Test, and at an ad-hoc visuomotor task evaluating manual visuomotor control. Secondary outcomes will include the scores at the Children's Hand Experience Questionnaire, Canadian Occupational Performance Measure, Melbourne Assessment of Unilateral Upper Limb Function, Gross Motor Function Measure, Vineland, Pediatric quality of life inventory. The evaluation points will include pre (T0), post (T1) and 3-month follow up (T2) assessments. Safety and tolerability will also be assessed. Results: The results of this trial will assess whether tVNS can effectively boost the effects of an intensive two-week bimanual training, in improving manual dexterity in children and adolescents with cerebral palsy, ensuring safety and tolerability throughout the intervention period.Clinical trial registration: ClinicalTrials.gov, NCT06372028.

PMID: 39220734

3. Effect of a Rehabilitation Program Including Home-Based Vibration-Assisted Therapy on Gait Parameters in Children with Cerebral Palsy

Stefanie Steven, Karoline Spiess, Bruno Lentzen, Nina Reinhart, Eckhard Schoenau, Ibrahim Duran

J Musculoskelet Neuronal Interact. 2024 Sep 1;24(3):259-266.

Objectives: The aim of the present study was to examine the effects of a rehabilitation program combined with a home-based vibration-assisted therapy on gait parameters in children with cerebral palsy (CP). Methods: In a retrospective study, 180 children, 101 boys and 79 girls, (mean age 7.2 ± 3.3 years) with CP at Gross Motor Function Classification System (GMFCS) Level I and Level II were examined using gait analyses with the Leonardo Mechanograph® Gangway at three measurement points. The measurements were conducted before (M0) and after a six-month rehabilitation period (M6), as well as 12 months after the commencement of rehabilitation (M12). The difference between measurement points M6-M0 (treatment interval) and M12-M6 (follow-up interval) were compared, and significance was determined using the Wilcoxon test. Results: Children with CP at GMFCS Level I and II demonstrated a significant improvement in gait efficiency (pathlength/distance M6-M0: -0.053 (SD 0.25) vs M12-M6: -0.008 (0.36), p=0.038). There were no significant difference in change of mean velocity and average step length between M6-M0 and M12-M6 (p=0.964 and p=0.611). Conclusions: The rehabilitation program seems to enhance gait efficiency in children with CP. German Clinical Trial Registry: DRKS0001131 at www.germanctr.de.

PMID: 39219323

4. Could Botox make walking easier in children with cerebral palsy?

No authors listed

Saudi Med J. 2024 Aug;45(9):1000.

No abstract available

PMID: 39218462

5. The impact of ankle-foot orthotics on selective motor control during gait in children and adolescents with cerebral palsy

Dan Keren, Marije Goudriaan, Shmuel Springer, Gilad Sorek, Nadia Dominici, Marjolein M van der Krogt, Annemieke I Buizer, Simon-Henri Schless

Gait Posture. 2024 Aug 24:114:48-54. doi: 10.1016/j.gaitpost.2024.08.079. Online ahead of print.

Background: Children with cerebral palsy (CP) often exhibit altered selective motor control during gait (SMCg). Ankle-foot-orthoses (AFOs) are used in this population to improve gait, by reducing the degrees of freedom at the ankle joint. However, the specific impact of AFOs on SMCg and whether this effect is more related to gait deviations or motor development remains unclear. Research question: Do AFOs impact SMCg, and is the change related to joint kinematics or age? Methods: Gait analysis data from 53 children and adolescents with spastic CP, walking both barefoot and with AFOs, were included. Electromyography data from six lower-limb muscles, and lower limb joint kinematics were analyzed for both walking conditions. SMCg was quantified by the total variance in electromyography activity accounted for by one synergy (tVAF1), where an increase in tVAF1 indicates a decrease in SMCg. Kinematic gait deviation was assessed using the Gait-Profile-Score (GPS) and sagittal plane ankle Gait-Variable-Score (ankle-GVS). All analyses were performed for the more clinically involved leg only. Results: Walking with AFOs resulted in a mean increase in tVAF1 of 0.02±0.07 (p=0.015) and a median increase in ankle-GVS of 3.4º (p>0.001). No significant changes were observed in GPS, and no correlation was found between the changes in tVAF1 and ankle-GVS. A significant positive moderate correlation was found between the change in tVAF1 and age, even with ankle-GVS as a covariate (r=0.45; p>0.001). Significance: Walking with an AFO resulted in a small decrease in SMCg, with large inter-participant variability. Younger participants showed a greater decrease in SMCg, which may indicate greater

neuromuscular plasticity in early developmental stages.

PMID: 39236422

6. Type and Distribution of Gross Motor Activity During Physical Therapy in Young Children With Cerebral Palsy

Laura A Prosser, Athylia C Paremski, Julie Skorup, Morgan Alcott, Samuel R Pierce

Phys Ther. 2024 Sep 3:pzae125. doi: 10.1093/ptj/pzae125. Online ahead of print.

Objective: Physical therapists routinely deliver and prescribe motor practice to improve function. The ability to select optimal practice regimens is limited by a current lack of detail in the measurement of motor practice. The objective of this study was to quantify the type, amount, and timing of gross motor practice during physical therapist sessions. Methods: A secondary video coding analysis of physical therapist sessions from the iMOVE clinical trial (NCT02340026) in young children with cerebral palsy (CP) was conducted. The 37 children who completed the treatment phase were included (mean age = 22.1 months). Children could initiate pulling to stand but were unable to walk. Videos of randomly selected therapy sessions were coded for gross motor activity (422 videos total). The 10 gross motor activity codes included lying, sitting, four point, crawling, kneeling, knee walking, standing, walking, transitions between floor postures, and transitions to/from an upright posture. Twenty percent of each video was double coded for reliability. Time per session, number of bouts, and median time per bout were calculated for each gross motor activity and for 2 aggregate measures: movement time and upright time. Results: Participants spent more than half of therapy time in sitting and standing combined (60.3%). Transitions occurred more frequently than any other motor activity (49.3 total transitions per session). Movement time accounted for 16.3% of therapy time. Upright time accounted for 53.3% of therapy time. Conclusions: Critical practice time to gain motor skill is not equivalent to chronological time or time spent in therapy. Toddlers with CP spent a small amount of therapy time moving. Future work should explore the relations between motor practice and rehabilitation outcomes. Impact: Physical therapists are ideally suited to detail the content of motor practice and ultimately to prescribe optimal patterns of motor practice. We report the characteristics of gross motor practice during therapy in children with CP.

PMID: 39223953

7. Expressive vocabulary intervention for four 2- to 3-year-old children with cerebral palsy and speech/language difficulties: A single-case A-B study

Anna Nyman, Carmela Miniscalco, Anette Lohmander, Sofia Strömbergsson

Int J Speech Lang Pathol. 2024 Sep 1:1-15. doi: 10.1080/17549507.2024.2373259. Online ahead of print.

Purpose: To evaluate a parent-delivered expressive vocabulary intervention using focused stimulation for young children with cerebral palsy (CP). Method: A single case A-B design was used. Use of expressive target words was evaluated during the baseline (A) and intervention (B) phases. Untrained expressive words were used as control behaviour. Four 2- to 3-year-old boys with CP and their parents participated. All participants had an expressive vocabulary of less than 50 words at study intake. Following training, parents delivered the intervention daily at home for 5 weeks. Generalisation of the intervention was measured with a parent-rated vocabulary checklist. Result: Two participants showed clear gains and one participant showed a smaller gain in target words. Two of these three participants did not improve on control words, but one participant increased his use of both target and control words. One participant did not increase his use of either control or target words. All participants improved on parent-reported expressive vocabulary, and for two children, improvements were large. Conclusion: Parent-delivered focused stimulation may lead to gains in expressive vocabulary in children with CP and speech and language difficulties. The intervention would be suitable for larger single-case studies with more experimental control.

PMID: 39218006

8. An instantaneous voice synthesis neuroprosthesis

Maitreyee Wairagkar, Nicholas S Card, Tyler Singer-Clark, Xianda Hou, Carrina Iacobacci, Leigh R Hochberg, David M Brandman, Sergey D Stavisky

bioRxiv [Preprint]. 2024 Aug 19:2024.08.14.607690. doi: 10.1101/2024.08.14.607690.

Brain computer interfaces (BCIs) have the potential to restore communication to people who have lost the ability to speak due to neurological disease or injury. BCIs have been used to translate the neural correlates of attempted speech into text1-3. However, text communication fails to capture the nuances of human speech such as prosody, intonation and immediately hearing one's own voice. Here, we demonstrate a "brain-to-voice" neuroprosthesis that instantaneously synthesizes voice with closed-loop audio feedback by decoding neural activity from 256 microelectrodes implanted into the ventral precentral gyrus of a man with amyotrophic lateral sclerosis and severe dysarthria. We overcame the challenge of lacking ground-truth speech for

training the neural decoder and were able to accurately synthesize his voice. Along with phonemic content, we were also able to decode paralinguistic features from intracortical activity, enabling the participant to modulate his BCI-synthesized voice in real-time to change intonation, emphasize words, and sing short melodies. These results demonstrate the feasibility of enabling people with paralysis to speak intelligibly and expressively through a BCI.

PMID: 39229047

9. Feasibility of a home-based home videogaming intervention with a family-centered approach for children with cerebral palsy: a randomized multiple baseline single-case experimental design

Daniela Chan-Víquez, Heilyn Fernández-Huertas, Carles Montserrat-Gonzalez, Ajmal Khan, Darcy Fehlings, Sarah Munce, F Virginia Wright, Elaine Biddiss

Randomized Controlled Trial J Neuroeng Rehabil. 2024 Sep 4;21(1):151. doi: 10.1186/s12984-024-01446-2.

Background: Worldwide, children with cerebral palsy (CP) living in underserved communities face barriers to accessing motor therapy services. This study assessed the implementation and effectiveness of an 8-week, upper limb (UL) home-based intervention with a movement-tracking videogame (Bootle Blast) in Costa Rican children with CP. Methods: Children established a weekly playtime goal and two UL activities of daily living (ADLs) that they would like to improve on. A multiple -baseline, single-case experimental design, was used with the Performance Quality Rating Scale (PQRS) as the repeated measure to track changes in performance of the selected ADLs between the baseline (usual care) and intervention (Bootle Blast) phases. The Canadian Occupational Performance Measure (COPM), the Box and Blocks Test (BBT) and the Children's Hand-Use Experience Questionnaire (CHEQ) were collected before and after the intervention. Technical barriers were documented during weekly video calls with a monitoring therapist. Treatment effect size, slope changes and percentage of non-overlapping data were identified for the PQRS. Descriptive statistics summarized results for the BBT, CHEQ, videogame logs (e.g., playtime) and technical barriers. Results: Fifteen children participated and 13 completed the intervention. Both participants who dropped out did so after completing baseline assessments, but before experiencing Bootle Blast. Children's mean active playtime (i.e., mini-games targeting the UL) across the 8-weeks was 377 min, while mean total time spent engaging with Bootle Blast (active + passive play time [e.g., time navigating menus, reviewing rewards]) was 728 min. In total, eight technical issues (from five children) were reported, and all but three were resolved within 48 h. Partial effectiveness was associated with the intervention. Specifically, 85% of participants improved on the PQRS and 69% achieved clinically important improvements ≥ 2 points in performance on the COPM. Children improved by 1.8 blocks on average on the BBT, while on the CHEQ, five children had a clinically important increase of 10% of the total number of UL activities performed with both hands. Conclusion: Bootle Blast is a feasible and effective option to facilitate access and engage children with cerebral palsy in UL home rehabilitation. Trial registration Trial registration number: NCT05403567.

PMID: 39227911

10. MR-guided focused ultrasound in pediatric neurosurgery: current insights, technical challenges, and lessons learned from 45 treatments at Children's National Hospital

Gregory F Keating, Kelsi M Chesney, Nirali Patel, Lindsay Kilburn, Adriana Fonseca, Roger J Packer, Chaitanya Challa, Patrick F O'Brien, Daniel A Donoho, John S Myseros, Chima Oluigbo, Robert F Keating, Hasan R Syed

Neurosurg Focus. 2024 Sep 1;57(3):E6. doi: 10.3171/2024.6.FOCUS24332.

Objective: MR-guided focused ultrasound (MRgFUS) is an evolving technology with numerous present and potential applications in pediatric neurosurgery. The aim of this study was to describe the use of MRgFUS, technical challenges, complications, and lessons learned at a single children's hospital. Methods: A retrospective analysis was performed of a prospectively collected database of all pediatric patients undergoing investigational use of MRgFUS for treatment of various neurosurgical pathologies at Children's National Hospital. Treatment details, clinical workflow, and standard operating procedures are described. Patient demographics, procedure duration, and complications were obtained through a chart review of anesthesia and operative reports. Results: In total, 45 MRgFUS procedures were performed on 14 patients for treatment of diffuse intrinsic pontine glioma (n = 12), low-grade glioma (n = 1), or secondary dystonia (n = 1) between January 2022 and April 2024. The mean age at treatment was 9 (range 5-22) years, and 64% of the patients were male. With increased experience, the total anesthesia time, sonication time, and change in core body temperature during treatment all significantly decreased. Complications affected 4.4% of patients, including 1 case of scalp edema and 1 patient with a postprocedure epidural hematoma. Device malfunction requiring abortion of the procedure occurred in 1 case (2.2%). Technical challenges related to transducer malfunction and sonication errors occurred in 6.7% and 11.1% of cases, respectively, all overcome by subsequent user modifications. Conclusions: The authors describe the largest series on MRgFUS technical aspects in pediatric neurosurgery at a single institution, comprising 45 total treatments. This study emphasizes potential technical challenges and provides valuable insights into the nuances of its application in pediatric patients.

PMID: 39217632

11. Fetal Intraparenchymal Hemorrhage Imaging Patterns, Etiology, and Outcomes: A Single Center Cohort Study

Rachel Vassar, Elizabeth George, Andrew Mogga, Yi Li, Mary E Norton, Orit Glenn, Dawn Gano

Ann Neurol. 2024 Aug 31. doi: 10.1002/ana.27072. Online ahead of print.

Objective: This study examines associations among fetal brain magnetic resonance imaging (MRI) injury patterns, etiologies, and outcomes in fetal intraparenchymal hemorrhage (IPH). Methods: This is a retrospective, single-center cohort study of IPH diagnosed on fetal MRI (1996-2022). IPH and associated abnormalities were categorized by 2 pediatric neuroradiologists; electronic medical records were reviewed by 2 pediatric neurologists to classify etiology and outcomes including cerebral palsy, epilepsy, developmental delay, and death. Results: Forty-four fetuses with IPH were identified (34 singleton and 10 twin gestations) with MRI at median 24 weeks gestation (interquartile range [IQR] = 22-28 weeks). IPH was commonly supratentorial (84%) and focal (50%) or focal with diffuse injury (43%) and was often associated with germinal matrix hemorrhage (GMH; 75%) and/or intraventricular hemorrhage (IVH; 52%). An etiology was identified in 75%, including twin-twin transfusion syndrome (TTTS, n = 10), COL4A1/2 variants (n = 8), or other fetal/maternal conditions (n = 15). COL4A1/2 variants were associated with focal IPH and the presence of hemorrhagic porencephaly, and intrauterine transfusion was associated with infratentorial hemorrhage. Twenty-two fetuses were liveborn, and 18 pregnancies were terminated. Among those with follow-up ≥ 12 months (median = 7 years), 12 of 13 had cerebral palsy, 6 of 13 had developmental delay, and 5 of 13 had epilepsy. Interpretation: An etiology for fetal IPH with or without GMH-IVH is identified in most cases in our cohort and is commonly TTTS, COL4A1/2 variants, or other maternal/fetal comorbidities. Pattern of fetal IPH on MRI is associated with etiology. Cerebral palsy and neurodevelopmental impairment were common in liveborn infants. Genetic studies should be considered in cases of fetal IPH without an otherwise apparent cause. ANN NEUROL 2024.

PMID: 39215698

12. Are important predictors of adverse outcome in children with symptomatic congenital cytomegalovirus infection overlooked in clinical settings?

Ivana Đaković, Ivica Kostović, Katarina Vulin, Iva Prvčić, Goran Tešović, Goran Krakar, Tomislav Gojmerac, Jadranka Sekelj Fureš, Vlatka Mejaški Bošnjak

Observational Study J Int Med Res. 2024 Sep;52(9):3000605241274226. doi: 10.1177/03000605241274226.

Objective: Congenital cytomegalovirus infection (cCMV) is a common, frequently unrecognized cause of childhood disability. The aim of the present study was to determine the symptoms that raise the suspicion of cCMV, define the neurodevelopmental outcomes, and assess their correlations. Methods: This longitudinal observational study comprised 78 children with symptomatic cCMV who underwent neuropediatric follow-up for 4 to 17.9 years. Results: Symptoms of central nervous system involvement, hearing/visual impairments, and hepatic involvement were mostly recognized. The average age of disease suspicion was 3.3 months. In terms of outcomes, 10.53% of the children developed complex minor neurological dysfunction and 23.68% developed cerebral palsy. Visual and hearing impairments occurred in 38.16% and 14.47% of patients, respectively. Intellectual disability was present in 30.26% of patients, and epilepsy in 21.05%. Microcephaly and hearing impairment was significantly associated with overall neurodevelopmental outcome. Microcephaly was also associated with poor motor outcomes, hearing impairment, and severe visual impairment. Furthermore, microcephaly and intrauterine growth restriction were significantly associated with poor cognitive outcomes. Conclusion: Symptoms that raised the suspicion of cCMV-especially microcephaly, hearing impairment, and intrauterine growth restriction-were important parameters that were associated with outcomes; however, their recognition was often insufficient and/or late.

PMID: 39224952

13. A randomized controlled trial protocol for the introduction of a multidisciplinary individualized nutritional intervention in children with cerebral palsy

Ruzha Pancheva, Stanka A Fitneva, Rositsa Chamova, Dimitar Marinov, Albena Toneva, Stanislava Hadzhieva, Rozalina Braykova, Nikoleta Yoncheva, Stefka Tsvetanova, Silviya Nikolova, Natalya Usheva, Koen Huysentruyt, Karina Dimova, Yana Bocheva, Stanislava Pavlova, Petya Hristanova

Contemp Clin Trials Commun. 2024 Aug 7:41:101343. doi: 10.1016/j.conctc.2024.101343. eCollection 2024 Oct.

Introduction: Children with Cerebral Palsy (CP) encounter substantial nutritional challenges that impair their health and quality of life. Despite the importance of nutrition in managing CP and the recognition of physiological, behavioral, and social causes of malnutrition, research on the effectiveness of individualized nutritional interventions developed and supported by multidisciplinary teams is scarce. Aim: The study will evaluate the impact of an individualized nutritional intervention developed and supported by a multidisciplinary team on the anthropometric outcomes and overall health of children with CP. Methods: A single-center, randomized controlled trial, conducted at the Medical University of Varna, Bulgaria, will enroll 100 children aged 2-12 years and diagnosed with CP. Participants will be randomly assigned to either an intervention group,

receiving comprehensive structured dietary assessment and individualized nutrition plan developed by a multidisciplinary team of experts, or to a standard care group. Outcomes assessed will focus on anthropometric measures of nutritional status, but also include health outcomes, child development and clinical assessments, and quality of life indicators. Ethics: Ethical approval for this study has been obtained from the Medical Ethics Committee at the Medical University of Varna (Protocol No. 134 dated 20.07.2023). Conclusion: This study will assess the benefits of a multidisciplinary, individualized nutritional intervention for children with CP. The findings will have implications for clinical guidelines and interventions aiming to improve their care and quality of life.

PMID: 39224169

14. Alpha oscillations during visual selective attention are aberrant in youth and adults with cerebral palsy

Rashelle M Hoffman, Michael P Trevarrow, Brandon J Lew, Tony W Wilson, Max J Kurz

Cereb Cortex. 2024 Sep 3;34(9):bhae365. doi: 10.1093/cercor/bhae365.

Our understanding of the neurobiology underlying cognitive dysfunction in persons with cerebral palsy is very limited, especially in the neurocognitive domain of visual selective attention. This investigation utilized magnetoencephalography and an Eriksen arrow-based flanker task to quantify the dynamics underlying selective attention in a cohort of youth and adults with cerebral palsy (n = 31; age range = 9 to 47 yr) and neurotypical controls (n = 38; age range = 11 to 49 yr). The magnetoencephalography data were transformed into the time-frequency domain to identify neural oscillatory responses and imaged using a beamforming approach. The behavioral results indicated that all participants exhibited a flanker effect (greater response time for the incongruent compared to congruent condition) and that individuals with cerebral palsy were slower and less accurate during task performance. We computed interference maps to focus on the attentional component and found aberrant alpha (8 to 14 Hz) oscillations in the right primary visual cortices in the group with cerebral palsy. Alpha and theta (4 to 7 Hz) oscillations were also seen in the left and right insula, and these oscillations varied with age across all participants. Overall, persons with cerebral palsy exhibit deficiencies in the cortical dynamics serving visual selective attention, but these aberrations do not appear to be uniquely affected by age.

PMID: 39233375

15. Lessons learnt from a 2017 Irish national clinical claims review: a retrospective observational study

Karen A Power, Irene O'Byrne Maguire, Noelle Byrne, Deirdre Walsh, Karen Robinson, Mark McCullagh, Yvonne Fallon, Mary Godfrey, Ann Duffy, Claire O'Regan, Mairead Twohig, Cathal O'Keeffe

Observational Study BMJ Open Qual. 2024 Sep 3;13(3):e002688. doi: 10.1136/bmjoq-2023-002688.

Objective: Learning from adverse outcomes in health and social care is critical to advancing a culture of patient safety and reducing the likelihood of future preventable harm to service users. This review aims to present an overview of all clinical claims finalised in one calendar year involving publicly funded health and social care providers in Ireland. Design: This is a retrospective observational study. The Clinical Risk Unit (CRU) of the State Claims Agency identified all service-user clinical claims finalised between 1 January 2017 and 31 December 2017 from Ireland's National Incident Management System (n=713). Claims that had incurred financial damages were considered for further analysis (n=356). 202 claims underwent an in-depth qualitative review. Of these, 57 related to maternity and gynaecology, 64 to surgery, 46 to medicine, 20 to community health and social care and 15 related to children's healthcare. Results: The services of surgery and medicine ranked first and second, respectively, in terms of a number of claims. Claims in maternity services, despite ranking third in terms of claims numbers, resulted in the highest claims costs. Catastrophic injuries in babies resulting in cerebral palsy or other brain injury accounted for the majority of this cost. Diagnostic errors and inadequate or substandard communication, either with service users and/or interprofessional communication with colleagues, emerged as common issues across all clinical areas analysed. Quantitative analysis of contributory factors demonstrated that the complexity and seriousness of the service user's condition was a significant contributory factor in the occurrence of incidents leading to claims. Conclusion: This national report identifies common issues resulting in claims. Targeting these issues could mitigate patient safety risks and reduce the cost of claims.

PMID: 39231573

16. Major structural congenital anomalies and causal pathways in people with cerebral palsy

Susan M Reid, Gina L Hinwood, Angela T Guzys, Rod W Hunt, Dinah S Reddihough

Dev Med Child Neurol. 2024 Sep 5. doi: 10.1111/dmcn.16073. Online ahead of print.

Aim: To determine the proportion of persons with cerebral palsy (CP) with major congenital anomalies, factors associated with the presence of anomalies, body systems involved, potential contribution to CP aetiology, and causal pathway subgroups

implicated. Method: This population-based, observational study involved a cohort of 2238 persons born in one Australian state between 1999 and 2017. Major congenital anomalies were classified as affecting cerebral, cardiac, or other body systems, with further categorization as single or multisystem. We determined the potential for anomalies to contribute to the development of CP across causal pathway subgroups that were broadly categorized as developmental or involving destructive brain insults. Results: Of persons with CP, 23% had major congenital anomalies and 17% of the cohort had anomalies that potentially contributed to the development of CP. Consistent with higher odds of parental consanguinity, maternal grand multiparity, and dysmorphic features in the group with anomalies, 82% of pathogenic anomalies, present in 14% of the cohort, were cerebral and involved developmental causal pathways. Only 3% (predominantly severe cardiac anomalies) were related to destructive brain insults. Interpretation: The study provides context for the impact on rates of CP of preventive measures or other changes in incidence or management of congenital anomalies.

PMID: 39233603

17. Causal pathways of cerebral palsy in individuals with congenital anomalies: A cardiologist's perspective

Mads Damkjær

Dev Med Child Neurol. 2024 Sep 5. doi: 10.1111/dmcn.16077. Online ahead of print.

No abstract available

PMID: 39233580

18. Development and Validation of the Questionnaire of Young People's Participation-Young Adults (QYPP-YA)

Chirine Cytera, Holger Muehlan, Marion Rapp, Audrey Guyard, Ute Thyen, Silke Schmidt, Henriette Markwart; TRANS-DISAB Collaborative Group

Child Care Health Dev. 2024 Sep;50(5):e13326. doi: 10.1111/cch.13326.

Background: The Questionnaire of Young People's Participation (QYPP) was developed for use in children and adolescents. To track participation throughout transition from childhood to adulthood, we adapted it for young adults using focus groups. Aim of this study was to validate this measure, the QYPP-Young Adults (QYPP-YA). Methods: We recruited young adults with cerebral palsy (CP) and a representative, same-aged sample of the general population (GP). The GP-sample was split into two equivalent subsamples, one part to identify the factor structure via exploratory factor analysis and another part to test the resulting model via confirmatory factor analysis. Reliability and different forms of validity were investigated. Results: The final QYPP-YA includes 17 items assigned to six domains (Autonomy, Independency, Intimate Relationships, Interpersonal Relationships, Social Life, Online Communication). Scales show satisfying internal consistencies in the CP-sample and in the GP-sample, except for 'Online Communication'. Convergent, divergent and known-group validity were confirmed. Conclusions: The QYPP-YA instrument features promising psychometric characteristics to assess key domains of participation in healthy and disabled young adults. It provides a multidimensional, economic and sound assessment for use in population surveys and clinical trials.

PMID: 39237254

19. Preparing Adults with Cerebral Palsy to Move from Assisted to Independent Living

Lori Rosenberg, Shira Zecharia, Yafit Gilboa, Anat Golos

Can J Occup Ther. 2024 Sep 5:84174241274743. doi: 10.1177/00084174241274743. Online ahead of print.

Background. To evaluate effectiveness of The Greenhouse for autonomy and independence to prepare adults with severe cerebral palsy (CP) for the transition from assisted to independent living. The intervention combines weekly individual sessions using Pathways and Resources for Engagement and Participation together with weekly group sessions. Method. Seven adults with severe CP ages 23-45 years (M = 35; SD = 10) participated in the 20-week program. An Interrupted Time Series quasi-experimental design was used, assessing the intervention effect over time. The Canadian Occupational Performance Measure was administered bi-weekly from baseline to post-intervention to assess activity performance. Functional Independence Measure (FIM), Wheelchair use Confidence scale (WheelCon) and Impact on Participation and Autonomy (IPA) were administered baseline (4 weeks pre-intervention), mid and post-intervention, using Freidman test. Interviews were conducted four weeks after intervention. Findings. All participants' activity performance improved over time with significant clinical improvement in 95% of COPM goals. Significant improvement was seen in FIM ($\chi 2 = 8.07$, $p = .018$) and WheelCon ($\chi 2 = 7.18$, $p = .028$) though not in IPA. Participants described being better prepared, however more aware of challenges. Conclusion. The findings suggest the program may be effective to help adults with severe CP attain goals related to independent living and enhance function.

PMID: 39238244

20. Individualized telehealth home programme for children with cerebral palsy during the COVID-19 pandemic

Rachel H S Oliveira, Marisa C Mancini, Priscilla R P Figueiredo, Leonardo C Abrahão, Edna A Reis, Andrew M Gordon, Marina B Brandão

Dev Med Child Neurol. 2024 Sep 6. doi: 10.1111/dmcn.16072. Online ahead of print.

Aim: To analyse the effects of an individualized telehealth home programme on the performance of functional goals of children and adolescents with cerebral palsy (CP) during the COVID-19 pandemic. Method: A prospective single-group intervention study with children/adolescents with CP (n = 144; median age = 92 months [Q1 = 44.0, Q3 = 148.8]; 74 males, 70 females), representing all Gross Motor Function Classification System (GMFCS) levels participated in a 4-month home programme in Brazil. An interdisciplinary team encouraged families to choose a functional goal to be trained. The Canadian Occupational Performance Measure (COPM) was used at pre-intervention (T1), post-intervention (T2), and 3-month follow-up (T3). The differences in COPM scores at T1, T2, and T3 were evaluated using Friedman's test. The effect size was calculated using Cohen's d. Univariate analysis was included. Results: Significant improvements were observed after the intervention, with maintenance of scores after 3 months ($p < 0.001$, dperformance = 1.33; dsatisfaction = 1.31). None of the tested variables (child's abilities, age, caregiver's educational level, perception of family-centredness, and type of goal) were significantly related to the change scores. Interpretation: The individualized remote telehealth home programme can be a potential intervention, especially for children with CP classified in GMFCS levels IV and V. Also, this intervention provided a possible solution to help some children and their families in performing prioritized functional goals during the pandemic period.

PMID: 39240105

21. Ablation of Iron Regulatory Protein 2 produces a neurological disorder characterized by motor, somatosensory, and executive dysfunction in mice

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Iron is an important cofactor for many proteins and is used to create Fe-S clusters and heme prosthetic groups that enzymes use to catalyze enzymatic reactions. Proteins involved in the import, export, and sequestration of iron are regulated by Iron Regulatory Proteins (IRPs). Recently, a patient with bi-allelic loss of function mutations in IREB2 leading to the absence of IRP2 protein was discovered. The patient failed to achieve developmental milestones and was diagnosed with dystonic cerebral palsy, epilepsy, microcytic hypochromic anemia, and frontal lobe atrophy. Several more IREB2 deficient patients subsequently identified manifested similar neurological problems. To better understand the manifestations of this novel neurological disease, we subjected an Irp2-null mouse model to extensive behavioral testing. Irp2-null mice had a significant motor deficit demonstrated by reduced performance on rotarod and hanging wire tests. Somatosensory function was also compromised in hot and cold plate assays. Their spatial search strategy was impaired in the Barnes maze and they exhibited a difficulty in flexibly adapting their response in the operant touchscreen reversal learning task. The latter is a cognitive behavior known to require an intact prefrontal cortex. These results suggest that loss of Irp2 in mice causes motor and behavioral deficits that faithfully reflect the IREB2 patient's neurodegenerative disorder.

PMID: 39239479

22. Different care mode alter composition and function of gut microbiota in cerebral palsy children

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Introduction: Specialized care is essential for the recovery of children with cerebral palsy (CP). This study investigates how different care modes impact the gut microbiota. Methods: Fecal samples from 32 children were collected, among whom those cared for by family (n = 21) were selected as the observation group, and those cared for by children's welfare institutions (n = 11) were selected as the control group (registration number of LGFYYXLL-024). The gut microbiota profiles were analyzed. Results: There was no significant difference in the α-diversity of the gut microbiota and the abundance at the phylum level. However, at the genus level, the observation group showed a significant increase in the abundance of butyrate-producing bacteria Bacteroides and Lachnospiracea incertae sedis ($P < 0.05$), and a significant decrease in the abundance of opportunistic pathogens Prevotella, Clostridium cluster IV, Oscillibacter, and Fusobacterium ($P < 0.05$). Additionally, lipid metabolism, carbohydrate metabolism, transcription, cellular processes and signaling, and membrane transport were significantly upregulated in the observation group. Lipid metabolism was positively correlated with Bacteroides and Lachnospiracea incertae sedis, indicating a positive impact of the family-centered care mode on bacterial metabolism processes. Discussion:

This study highlights that the family-centered care mode had a positive impact on the composition and function of the gut microbiota. The study provides valuable insights into the relationship between care mode and gut microbiota, which can inspire the development of interventions for cerebral palsy.

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23. Tyrosine hydroxylase deficiency in a child initially misdiagnosed as cerebral palsy combined with epilepsy

Xiaoli Qi, Jing Zhang, Zhiqiang Sheng

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No abstract available

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