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## Interventions and Management

### 1. Pelvis radiographs in children with cerebral palsy: effects of patient positioning on calculating migration percentages

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Pediatr Radiol. 2023 Oct 14. doi: 10.1007/s00247-023-05783-7. Online ahead of print.

**Background:** Hip displacement in children with cerebral palsy (CP) is monitored by measuring migration percentage on anteroposterior pelvis radiographs. However, proper positioning for radiography in children with spasticity is difficult. The reliability and accuracy of migration percentage as a function of patient positioning is unknown. **Objective:** To determine the effects of patient positioning on migration percentage measurements in children with CP. **Materials and methods:** We identified children with CP ( $\leq 18$ -year-old) with pelvis CT and anteroposterior pelvis radiograph obtained  $< 6$  months apart (10/2018-11/2021). Digitally reconstructed radiographs were generated from each pelvis CT, to simulate nine different patient positions: neutral;  $10^\circ$  and  $20^\circ$  lordosis and kyphosis; and  $10^\circ$  and  $20^\circ$  right rotation and left rotation. Two radiologists measured migration percentages from the simulated and real pelvis radiographs. We used Spearman's rho to assess inter-rater reliability, and Wilcoxon signed rank test to determine statistical significance. **Results:** We studied sixty-three children (male=41; median age=8 years; range=4-18 years). The two radiologists' migration percentage measurements were highly correlated with each other across all simulated and real radiographs (Spearman's rho=0.86-0.99,  $P < 0.01$ ). For both readers and hips, migration percentages measured from real radiographs were significantly different from those measured from neutral simulated radiographs ( $P < 0.01$ ), with median absolute difference=5-6 percentage points (PP) and interquartile range (IQR)=9-12 PP. When comparing migration percentage measurements from neutral simulated radiographs to those in kyphosis/lordosis and right/left rotations, median absolute differences were 2-4 PP (IQR=3-8 PP) and 4-15 PP (IQR=6-17 PP), respectively. **Conclusion:** Inter-rater reliability of measured migration percentages is high, but accuracy decreases with patient positional changes.

PMID: [37833504](https://pubmed.ncbi.nlm.nih.gov/37833504/)

### 2. Functional electrical stimulation during walking in children with unilateral spastic cerebral palsy: A randomized cross-over trial

Irene Moll, Rik G J Marcellis, Sabine M Fleuren, Marcel L P Coenen, Rachel H J Senden, Paul J B Willems, Lucianne A W M Speth, M Adhiambo Witlox, Kenneth Meijer, R Jeroen Vermeulen

Dev Med Child Neurol. 2023 Oct 12. doi: 10.1111/dmcn.15779. Online ahead of print.

**Aim:** To study if functional electrical stimulation (FES) of the peroneal nerve, which activates dorsiflexion, can improve body functions, activities, and participation and could be an effective alternative treatment in individuals with unilateral spastic cerebral palsy (CP). **Method:** A randomized cross-over trial was performed in 25 children with unilateral spastic CP (classified in Gross Motor Function Classification System levels I and II) aged 4 to 18 years (median age at inclusion 9 years 8 months, interquartile range = 7 years-13 years 8 months), 15 patients were male. The study consisted of two 12-week blocks of treatment, that is, conventional treatment (ankle foot orthosis [AFO] or adapted shoes) and FES, separated by a 6-week

washout period. Outcome measures included the Goal Attainment Scale (GAS), the Cerebral Palsy Quality of Life questionnaire, and a three-dimensional gait analysis. Results: Eighteen patients completed the trial. The proportion of GAS goals achieved was not significantly higher in the FES versus the conventional treatment phase (goal 1  $p = 0.065$ ; goal 2  $p = 1.00$ ). When walking while stimulated with FES, ankle dorsiflexion during mid-swing decreased over time ( $p = 0.006$ , average decrease of  $4.8^\circ$  with FES), with a preserved increased ankle range of motion compared to conventional treatment ( $p < 0.001$ , mean range of motion with FES  $+10.1^\circ$  compared to AFO). No changes were found in the standard physical examination or regarding satisfaction with orthoses and feelings about the ability to dress yourself. In four patients, FES therapy failed; in 12 patients FES therapy continued after the trial. Interpretation: FES is not significantly worse than AFO; however, patient selection is critical, and a testing period and thorough follow-up are needed.

PMID: [37823431](#)

### **3. Feasibility of using the NewGait assistive device for correcting gait deviations in individuals with various neurological disorders: Case study**

Ksenia I Ustinova, Joseph E Langenderfer

Case Reports Physiother Res Int. 2023 Oct 11:e2055. doi: 10.1002/pri.2055. Online ahead of print.

Purpose: Impaired gait is one of the earliest, most devastating, and long-lasting symptoms associated with neurological disorders. This study tested the feasibility of wearing the NewGait rehabilitative device in individuals with gait impairments due to the most common neurological disorders. Methods: Seven participants with gait impairments due to strokes, Multiple Sclerosis, peripheral neuropathies, Cerebral Palsy (CP) and Parkinson's Disease (PD) were included in the study. Their walking with and without wearing the NewGait was analyzed and compared using the Vicon T160 system for motion analysis. Gait velocity, step length, foot clearance, lateral displacement of the Center of Mass, gait deviation and symmetry indexes were compared using two standard deviation band method for each participant. Results: Participants subjectively assessed the NewGait as a comfortable device to wear and showed immediate gait improvements to varying degrees. Most improvements were observed in participants with muscle weakness due to peripheral neuropathies, stroke, MS, and CP. These participants improved their foot clearance, gait velocity, and step length. Participants with cerebellar stroke and PD increased their gait stability. All participants demonstrated a reduction in composite gait deviation indexes. Not all gait parameters, though, showed immediate changes. Conclusion: The results suggest that the NewGait rehabilitative device is feasible and useful for correcting gait impairments caused by neurological deficits. Participants may need to wear this device for longer periods of time in order to achieve long lasting changes in the gait pattern, rather than an immediate correction.

PMID: [37818770](#)

### **4. Effects of a multi-component virtual reality program on motor skills and functional postural control in children with hemiplegic cerebral palsy**

Meysam Roostaei, Marzieh Babae, Seyedmostafa Alavian, Narjes Jafari, Seyed Mansoor Rayegani, Saeed Behzadipour

Heliyon. 2023 Sep 9;9(9):e19883. doi: 10.1016/j.heliyon.2023.e19883. eCollection 2023 Sep.

Background: Pediatric neurorehabilitation has recently employed virtual reality (VR) technologies as a platform to design and implement novel modalities. Aims: To evaluate the feasibility of a multi-component VR-based program on motor skills and functional postural control for children with hemiplegic cerebral palsy (HCP). Methods: A single-case-experimental design was conducted on eight children with HCP ( $12.33 \pm 4.71$  years and GMFCS= II, I). The VR-based program consisted of 3 sessions per week for four weeks. Timed Up and Go (TUG) test, Functional Reach Test (FRT), Pediatric Balance Scale (PBS), Activities Scale for Kids (ASK), ABILHAND-Kids, and Box and Block Test (BBT) were used to evaluate functional changes. Outcomes and results: Statistical analysis showed that improvements in functional postural control were significant on at least one balance measure for seven out of eight participants during the intervention phase. For all participants, a significant increase was observed in the BBT scores. Before-after intervention analysis revealed statistically significant improvements in PBS ( $z = -2.52$ ,  $p \leq 0.01$ ), ABILHAND-Kids ( $z = -2.25$ ,  $p \leq 0.01$ ), and ASK ( $z = -2.38$ ,  $p \leq 0.01$ ). Conclusions and implications: This study provided early evidence of the effectiveness of the multi-component VR-based program in children with HCP. However, future studies with randomized controlled trial design are needed to evaluate the long-term effects and compare them with conventional rehabilitation practice.

PMID: [37809784](#)

### **5. Comparison of biomechanical analysis results using different musculoskeletal models for children with cerebral palsy**

Zhibo Jing, Jianda Han, Juanjuan Zhang

Front Bioeng Biotechnol. 2023 Sep 26;11:1217918. doi: 10.3389/fbioe.2023.1217918. eCollection 2023.

**Introduction:** Musculoskeletal model-based simulations have gained popularity as a tool for analyzing human movement biomechanics. However, when examining the same gait, different models with varying anatomical data and assumptions may produce inconsistent biomechanical results. This inconsistency is particularly relevant for children with cerebral palsy, who often exhibit multiple pathological gait patterns that can impact model outputs. **Methods:** The aim of this study was to investigate the effect of selecting musculoskeletal models on the biomechanical analysis results in children with cerebral palsy. Gait data were collected from multiple participants at slow, medium, and fast velocities. Joint kinematics, joint dynamics, and muscle activation were calculated using six popular musculoskeletal models within a biomechanical simulation environment. **Results:** The degree of inconsistency, measured as the root-mean-square deviation, in kinematic and kinetic results produced by the different models ranged from 4% to 40% joint motion range and 0%-28% joint moment range, respectively. The correlation between the results of the different models (both kinematic and kinetic) was good ( $R > 0.85$ ,  $P < 0.01$ ), with a stronger correlation observed in the kinetic results. Four of the six models showed a positive correlation between the simulated muscle activation of rectus femoris and the surface EMG, while all models exhibited a positive correlation between the activation of medial gastrocnemius and the surface EMG ( $P < 0.01$ ). **Discussion:** These results provide insights into the consistency of model results, factors influencing consistency, characteristics of each model's outputs, mechanisms underlying these characteristics, and feasible applications for each model. By elucidating the impact of model selection on biomechanical analysis outcomes, this study advances the field's understanding of musculoskeletal modeling and its implications for clinical gait analysis model decision-making in children with cerebral palsy.

PMID: [37823025](#)

## 6. An implantable, wireless, battery-free system for tactile pressure sensing

Lin Du, Han Hao, Yixiao Ding, Andrew Gabros, Thomas C E Mier, Jan Van der Spiegel, Timothy H Lucas, Firooz Aflatouni, Andrew G Richardson, Mark G Allen

Microsyst Nanoeng. 2023 Oct 11;9:130. doi: 10.1038/s41378-023-00602-3. eCollection 2023.

The sense of touch is critical to dexterous use of the hands and thus an essential component of efforts to restore hand function after amputation or paralysis. Prosthetic systems have addressed this goal with wearable tactile sensors. However, such wearable sensors are suboptimal for neuroprosthetic systems designed to reanimate a patient's own paralyzed hand. Here, we developed an implantable tactile sensing system intended for subdermal placement. The system is composed of a microfabricated capacitive pressure sensor, a custom integrated circuit supporting wireless powering and data transmission, and a laser-fused hermetic silica package. The miniature device was validated through simulations, benchtop assessment, and testing in a primate hand. The sensor implanted in the fingertip accurately measured applied skin forces with a resolution of 4.3 mN. The output from this novel sensor could be encoded in the brain with microstimulation to provide tactile feedback. More broadly, the materials, system design, and fabrication approach establish new foundational capabilities for various applications of implantable sensing systems.

PMID: [37829157](#)

## 7. Dietary Intake, Feeding Pattern, and Nutritional Status of Children with Cerebral Palsy in Rural Bangladesh

Israt Jahan, Risad Sultana, Mousume Afroz, Mohammad Muhit, Nadia Badawi, Gulam Khandaker

Nutrients. 2023 Sep 29;15(19):4209. doi: 10.3390/nu15194209.

(1) **Background:** Data on immediate causes of malnutrition among children with Cerebral Palsy (CP) are limited in low- and middle-income countries (LMICs). We aimed to assess the dietary intake pattern, feeding characteristics, and nutritional status of children with CP in Bangladesh; (2) **Methods:** We conducted a descriptive observational study in Shahjampur, Bangladesh. Children with CP registered into the Bangladesh CP Register were included. Socio-demographic, clinical, dietary intake, feeding, gastro-intestinal conditions, and anthropometric data were collected. Descriptive and inferential statistics were reported; (3) **Results:** 75 children (mean (SD) age 3.6 (2.7) years, 42.7% female) and their caregivers participated. Overall, 53.6% and 46.4% of children were underweight and stunted, respectively. Two-thirds children consumed  $\leq 4$  out of 8 food groups. Meat, poultry, and fish; dairy products; and sugar consumption was lower among underweight children (43.4%, 48.8%, 25.0%) than others (56.7%, 51.2% 75.0%). Inappropriate feeding position was observed in 39.2% of children. Meal duration was  $>30$  min/meal for 21.7–28.0% children. Among all, 12.0% had feeding difficulties, 88.0% had  $\geq 1$  gastro-intestinal conditions; (4) **Conclusions:** The study reports preliminary data on the feeding characteristics, dietary intake, and nutritional status of children with CP in rural Bangladesh. The findings are crucial for cost-effective interventions, prevention, and management of malnutrition among children with CP in Bangladesh and other LMICs.

PMID: [37836493](#)

## 8. Oral Streptococcus mutans load among Indian children with cerebral palsy

Aniruddh Gandhi, Subhash Sonkesriya, Shovan Roy, Raman Mishra, Jatin Arora, Vineet Soni

Bioinformation. 2023 Feb 28;19(2):215-220. doi: 10.6026/97320630019215. eCollection 2023.

The motor impairments of cerebral palsy (CP) are typically accompanied by subsequent musculoskeletal issues, seizures, and abnormalities of sensation, intelligence, communication, and behaviour. These kids have a lower capacity for regulating oral health because of their poor voluntary movements. Poor oral hygiene brought on by insufficient brushing and flossing, increased use of sugary foods, and orally administered drugs puts people at risk for periodontal disorders and dental caries. Poor dental health and rising therapy demands establish a sadistic cycle that affects patient overall health and wellbeing. The purpose of this investigation was comparing kids with CP against healthy kids of comparable age group and demographic situation in order to evaluate status of oral health, current caries behavior using measurement of Streptococcus mutans concentrations in saliva, and treatment required. 204 study participants were divided into two categories: Category A and category B. Both categories consisted of 102 study participants. Category A consisted of study participants having CP while category B consisted of healthy normal controls with same age of same demographic features. Malocclusion, trauma, DMFS/ defs, gingival index, and Oral hygiene score (OHI), and were recorded for oral examinations of all study participants. However, no radiological assistance was utilized since minimal patient compliance existed in CP patients. When compared with the control category, the CP category had a higher detection of the DMFS index in the permanent teeth. The estimated defs for the CP category did not differ noticeably from the control category. In the CP category, status of hygiene of oral cavity was discovered to be substantially subpar. In comparison to the control category, the gingival condition of the CP category was noticeably worse. Treatment requirements were seen to require greater preventative care in the control category while, stainless steel crowns, pulpectomy and extractions were needed in the CP category. S. mutans was found in high concentrations in the salivary specimens of the CP category compared to the control category, indicating active dental caries and greater probability of further development.

PMID: [37814684](#)

## 9. Characteristics of Children with Cerebral Palsy and Their Utilization of Services in Saudi Arabia

Maysoun Nimer Saleh, Ahmad Alharbi, Abdulaziz Albalwi, Salem Alatawi, Maaidah Algamdi, Abdulaziz Alshahrani, Basil Al Bakri, Nihad Almasri

Healthcare (Basel). 2023 Oct 7;11(19):2690. doi: 10.3390/healthcare11192690.

The recent emergence of research on cerebral palsy (CP) in developing countries aims to improve knowledge on affected children and the utilization of the available services. This study seeks to describe children with CP in Saudi Arabia and service utilization as per Gross Motor Function Classification System (GMFCS) levels and geographic regions. A cross-sectional survey of 227 children with CP (Mean age 6.3, SD 3.9 years) was conducted. Parents reported on children's demographics, impairments, and service utilization. Half of the children (n = 113, 49.8%) had  $\geq 3$  impairments with speech, visual and learning impairments being the most frequent. The total number of impairments differed significantly by GMFCS,  $F(4, 218) = 8.87, p < 0.001$ . Most of the children (n = 86, 83.4%) used 2-5 services. Moreover, 139 (62.3%) did not attend school, 147 (65.9%) did not receive occupational therapy, and only 32 (14.3%) received speech therapy. More children in GMFCS level I did not receive neurologist services. Profiles of children and services were described by GMFCS and by regions. This was the first study to describe children with CP and service utilization in Saudi Arabia. Although many impairments affected the children, there was low utilization of related services. Data on service utilization and on unmet needs support a comprehensive approach to rehabilitation and the proper service allocation.

PMID: [37830727](#)

## 10. Chronic fatigue syndrome in caregivers of children with cerebral palsy and affecting factors

Tugce Pasin, Bilinc Dogruoz Karatekin, Ozge Pasin

North Clin Istanbul. 2023 Sep 22;10(5):642-650. doi: 10.14744/nci.2023.53533. eCollection 2023.

**Objective:** In this study, the frequency of chronic fatigue syndrome (CFS), sleep disturbances, and quality of life levels in mothers of children with cerebral palsy (CP) was compared in relation to the functional status of the child. **Methods:** The caregivers were evaluated with the sociodemographic data form, Chalder fatigue scale (ChFS), Fatigue Severity Scale (FSS), Pittsburgh Sleep Quality Index, and Short Form-12, respectively. In addition, the functional status of the child with CP was evaluated with the gross motor function classification system, manual ability classification system (MACS), communication function classification system, and eating and drinking ability classification system. **Results:** According to CDC-1994 criteria, 80.4% of the participating mothers have CFS (n=45). While the mean ChFS and FSS scores of housewives were found to be significantly higher than those of full-time workers ( $p=0.002$ ;  $p=0.003$ , respectively), the mean SF-12 MCS was found to be significantly lower ( $p=0.007$ ). The rate of housewives was found to be significantly higher in those diagnosed with CFS

( $p < 0.001$ ). The relationship between independent variables and dependent variables data sets as a result of canonical correlation analysis was obtained as 0.815. While the variable with the highest effect among the independent variables is the MACS variable, the variable with the highest percentage of explanation for the dependent variables is ChFS. Conclusion: The frequency of CFS is very high in mothers of children with CP, and the most important factors in the presence and severity of CFS are the mother's occupational status and the child's manual skills.

PMID: [37829755](#)

### **11. Tocolysis after preterm prelabor rupture of membranes and 5-year outcomes: a population-based cohort study (EPIPAGE-2)**

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Am J Obstet Gynecol. 2023 Oct 10:S0002-9378(23)00739-1. doi: 10.1016/j.ajog.2023.10.010. Online ahead of print.

**Background:** The administration of tocolysis after preterm prelabor rupture of membranes remains a controversial practice. In theory, reducing uterine contractility should delay delivery, and allow for optimal antenatal management, thereby reducing the risks of prematurity and adverse consequences over the life course. However, tocolysis may be associated with neonatal death or long-term adverse neurodevelopmental outcomes, mainly related to prolonged fetal exposure to intrauterine infection/inflammation. In a previous study, we showed that tocolysis administration was not associated with short-term benefits. There are currently no data available to evaluate the impact of tocolysis on neurodevelopmental outcomes in school-aged children born preterm in this clinical setting. **Objective:** To investigate whether tocolysis administered after preterm prelabor rupture of membranes is associated with neurodevelopmental outcomes at 5.5 years of age. **Study design:** We used data from a prospective population-based cohort study of preterm births recruited in 2011 (EPIPAGE-2), with a comprehensive medical and neurodevelopmental assessment at age 5.5 years. We included pregnant individuals with preterm prelabor rupture of membranes at 24-32 weeks in singleton pregnancies with a fetus alive at the time of rupture, birth at 24-34 weeks, and participation in assessment at 5.5 years of age. Exposure was the administration of any tocolytic treatment after preterm prelabor rupture of membranes. The main outcome was survival without moderate-to-severe neurodevelopmental disabilities at 5.5 years. Secondary outcomes included survival without any neurodevelopmental disabilities, cerebral palsy, full scale intelligence quotient, developmental coordination disorders, and behavioural difficulties. A propensity-score analysis was used to minimize the indication bias in estimating the treatment effect on outcomes. **Results:** Overall, 596/803 pregnant individuals (73.4%) received tocolysis after preterm prelabor rupture of membranes. At the 5.5-year follow-up, 82.7% and 82.5% of the children in the tocolysis and no tocolysis groups were alive without moderate-to-severe neurodevelopmental disabilities; 52.7% and 51.1% were alive without any neurodevelopmental disabilities. After applying multiple imputations and inverse probability of treatment weighting, we found no association between exposure to tocolysis and survival without moderate-to-severe neurodevelopmental disabilities (Odds Ratio 0.93; 95% confidence interval 0.55;1.60), survival without any neurodevelopmental disabilities (Odds Ratio 1.02; 95% confidence interval 0.65;1.61), or any of the other outcomes. **Conclusion:** There was no difference in neurodevelopmental outcomes at age 5.5 among children with and without antenatal exposure to tocolysis after preterm prelabor rupture of membranes. To date, its health benefits remain unproven, both in the short- and long-term.

PMID: [37827270](#)

### **12. Fluoroscopic-Assisted Tongue Suspension: Advancement and Innovation in the Management of Complex Pediatric Obstructive Sleep Apnea**

Taher Valika

Laryngoscope. 2023 Oct 12. doi: 10.1002/lary.31107. Online ahead of print.

**Objectives:** The primary aim of this study is to describe a novel surgical technique developed for tongue base suspension (TBS). The second aim of this study is to assess the efficacy of the developed procedure by quantifying preoperative and postoperative polysomnographic outcomes for pediatric patients undergoing fluoroscopic-assisted tongue suspension (FATS) with the Encore System. Our hypothesis is that our FATS technique will provide at least a 50% reduction in the Apnea/Hypopnea Index (AHI), including in the medically complex pediatric population. **Study design:** Retrospective case series. **Methods:** An electronic medical record review was conducted of patients who underwent FATS by a single surgeon at a tertiary care medical center between December 2019 and June 2022. Inclusion criteria included all patients <18 years old with evidence of OSA or sleep-disordered breathing and who had glossoptosis on sleep endoscopy. Data extracted from the medical record included age, gender, medical comorbidity history, reason for referral, history of airway surgeries, length of hospital stay, surgical complications data, and preoperative and postoperative polysomnographic data. Surgical success was defined by at least a 50% reduction in AHI. **Results:** Thirty patients (53.3% male) with a mean age of 6.3 ( $\pm 5.3$ , 0.16-17) years underwent FATS over the study period. Most patients (93%) had an underlying comorbidity: cerebral palsy (37%), chromosomal abnormalities (23%), Down syndrome (13%), Pierre-Robin sequence (10%), and obesity (10%). The majority of patients (77%)



were explicitly referred for tracheostomy placement secondary to failed management of OSA. 21 patients completed both preoperative and postoperative polysomnograms. The mean preoperative AHI, obstructive AHI (oAHI), and respiratory disturbance index (RDI) were 28.8 ( $\pm 19.8$ ), 30.8 ( $\pm 19.6$ ), and 30.5 ( $\pm 19.3$ ), respectively. The mean postoperative AHI, oAHI, and RDI were 7.3 ( $\pm 9.2$ ), 7.5 ( $\pm 9.1$ ), and 7.9 ( $\pm 9.3$ ), respectively. The mean change in AHI was -21.5 ( $\pm 21.4$ ) events/h ( $p < 0.01$ , 95% CI -29.0 to -11.4 events/h). The mean percentage decrease in AHI was 74.7%. The mean change in oAHI and RDI were -23.3 ( $\pm 21.9$ ) events/h ( $p < 0.01$ , 95% CI -39.9 to -21.4 events/h) and -22.5 ( $\pm 21.5$ ) events/h ( $p < 0.01$ , 95% CI -31.5 to -12.4 events/h), respectively. The mean percentage decrease in oAHI and RDI was 75.6% and 73.8%, respectively. Surgical success occurred in 16 of the 21 (76%) patients. Of the 23 patients referred explicitly for tracheostomy placement, 21 (91%) were able to avoid tracheostomy placement secondary to improvements in OSA. Conclusions: Patients undergoing fluoroscopic-assisted TBS revealed statistically significant improvements in AHI, oAHI, and RDI, with an overall surgical success rate of 76%. Complication rates were minimal, despite the complex nature of the study population. FATS should be considered a viable surgical approach in pediatric patients with an identified base of tongue obstruction and OSA.

PMID: [37823584](#)

### 13. Doxapram versus placebo in preterm newborns: a study protocol for an international double blinded multicentre randomized controlled trial (DOXA-trial)

Jarinda A Poppe, Robert B Flint, Anne Smits, Sten P Willemsen, Kelly K Storm, Debbie H Nuytemans, Wes Onland, Marten J Poley, Willem P de Boode, Katherine Carkeek, Vincent Cassart, Luc Cornette, Peter H Dijk, Marieke A C Hemels, Isabelle Hermans, Matthias C Hütten, Dorotya Kelen, Ellen H M de Kort, André A Kroon, Julie Lefevre, Katleen Plaskie, Breanne Stewart, Michiel Voeten, Mirjam M van Weissenbruch, Olivia Williams, Inge A Zonnenberg, Thierry Lacaze-Masmonteil, Arjan B Te Pas, Irwin K M Reiss, Anton H van Kaam, Karel Allegaert, G Jeroen Hutten, Sinno H P Simons

Trials. 2023 Oct 10;24(1):656. doi: 10.1186/s13063-023-07683-5.

Background: Apnoea of prematurity (AOP) is one of the most common diagnoses among preterm infants. AOP often leads to hypoxemia and bradycardia which are associated with an increased risk of death or disability. In addition to caffeine therapy and non-invasive respiratory support, doxapram might be used to reduce hypoxemic episodes and the need for invasive mechanical ventilation in preterm infants, thereby possibly improving their long-term outcome. However, high-quality trials on doxapram are lacking. The DOXA-trial therefore aims to investigate the safety and efficacy of doxapram compared to placebo in reducing the composite outcome of death or severe disability at 18 to 24 months corrected age. Methods: The DOXA-trial is a double blinded, multicentre, randomized, placebo-controlled trial conducted in the Netherlands, Belgium and Canada. A total of 396 preterm infants with a gestational age below 29 weeks, suffering from AOP unresponsive to non-invasive respiratory support and caffeine will be randomized to receive doxapram therapy or placebo. The primary outcome is death or severe disability, defined as cognitive delay, cerebral palsy, severe hearing loss, or bilateral blindness, at 18-24 months corrected age. Secondary outcomes are short-term neonatal morbidity, including duration of mechanical ventilation, bronchopulmonary dysplasia and necrotising enterocolitis, hospital mortality, adverse effects, pharmacokinetics and cost-effectiveness. Analysis will be on an intention-to-treat principle. Discussion: Doxapram has the potential to improve neonatal outcomes by improving respiration, but the safety concerns need to be weighed against the potential risks of invasive mechanical ventilation. It is unknown if the use of doxapram improves the long-term outcome. This forms the clinical equipoise of the current trial. This international, multicentre trial will provide the needed high-quality evidence on the efficacy and safety of doxapram in the treatment of AOP in preterm infants.

PMID: [37817255](#)

### 14. Association of EEG Background and Neurodevelopmental Outcome in Neonates With Hypoxic-Ischemic Encephalopathy Receiving Hypothermia

Hannah C Glass, Adam L Numis, Bryan A Comstock, Fernando F Gonzalez, Ulrike Mietzsch, Sonia Lomeli Bonifacio, Shavonne Massey, Cameron Thomas, Niranjana Natarajan, Dennis E Mayock, Gregory M Sokol, Krisa P Van Meurs, Kaashif A Ahmad, Nathalie Maitre, Patrick J Heagerty, Sandra E Juul, Yvonne W Wu, Courtney J Wusthoff

Neurology. 2023 Oct 10;10.1212/WNL.000000000207744. doi: 10.1212/WNL.000000000207744. Online ahead of print.

Background and objectives: Predicting neurodevelopmental outcome for neonates with hypoxic-ischemic encephalopathy (HIE) is important for clinical decision-making, care planning, and parent communication. We examined the relationship between EEG background and neurodevelopmental outcome among children enrolled in a trial of erythropoietin (Epo) or placebo for neonates with HIE treated with therapeutic hypothermia. Methods: Participants had EEG recorded throughout hypothermia. EEG background was classified as normal, discontinuous, or severely abnormal (defined as burst suppression, low voltage suppressed, or status epilepticus) at five one-hour epochs: onset of recording, 24, 36, 48, and 72 hours after birth. The predominant background pattern during the entire cEEG recording was calculated using the arithmetic mean of the five EEG background ratings (normal=0; discontinuous=1; severely abnormal=2) as follows: "predominantly normal" (mean=0), "normal/discontinuous" ( $0 < \text{mean} < 1$ ), "predominantly discontinuous" (mean=1), "discontinuous/severely abnormal" ( $1 < \text{mean} < 2$ ), or "predominantly severely abnormal" (mean=2). Primary outcome was death or neurodevelopmental

impairment (NDI) defined as cerebral palsy, Gross Motor Function Classification Score  $\geq 1$  or cognitive score  $< 90$  on Bayley Scales of Infant Toddler Development, 3rd edition at age two years. Neurodevelopment was also categorized into a 5-level ordinal measure: no, mild, moderate, or severe NDI, or death for secondary analysis. We used generalized linear regression models with robust standard errors to assess the relative risk of death or NDI by EEG background in both unadjusted and adjusted analyses controlling for effects of treatment group, sex, HIE severity, and study recruitment site. Results: Among 142 neonates, predominant background EEG pattern was predominantly normal in 35 (25%), normal/discontinuous in 68 (48%), predominantly discontinuous in 11 (7.7%), discontinuous/severely abnormal in 16 (11%), and predominantly severely abnormal in 12 (8.5%). Increasing severity of background across monitoring epochs was associated with increasingly worse clinical outcomes. Children with severe EEG background abnormality at any time point ( $n=36$ , 25%) were significantly more likely to die or have severe neurodevelopmental impairment at two years (adjusted relative risk: 7.95, 95% confidence interval 3.49-18.12). Discussion: EEG background is strongly associated with NDI at age two years. These results can be used to assist health care providers to plan follow-up care and counsel families for decision making related to goals of care. NCT02811263.

PMID: [37816642](#)

### 15. Nutritional status as predictors for quality of life among caregivers of children with severe cerebral palsy

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Transl Pediatr. 2023 Sep 18;12(9):1601-1618. doi: 10.21037/tp-23-195. Epub 2023 Sep 14.

Background: Individuals with severe cerebral palsy (CP) often experience various health issues, including feeding difficulties, which can adversely affect their nutritional status and caregivers' quality of life, e.g., more time spent for feeding rather than own selfcare. This study aimed to determine the prevalence of poor nutritional status among individuals with severe CP and explore its role as predictors for caregivers' quality of life. Methods: This cross-sectional study was conducted in a government hospital (Cheras Rehabilitation Hospital), Community-Based Rehabilitation (CBR) Program, and Spastic Centre [non-profit organization (NGO)] in Klang Valley, Malaysia. Seventy-one participants with Gross Motor Function Classification System (GMFCS) level IV and V were recruited. Sociodemographic data, health-associated data, and anthropometric data were collected. Caregivers' quality of life was assessed using the Pediatric Quality of Life Inventory (PedsQL) CP module version 3.0 which included domains such as Movement and Balance, Eating Activities, and School Activities. Results: Most participants were males (60.6%), with 61 classified as level IV in the GMFCS classification. According to the Eating and Drinking Ability Classification System (EDACS), 59 children were at level IV. In total, 40.8% were underweight according to CP growth chart and 49.3% had mid-upper arm circumference (MUAC) readings below the 5th percentile. The lowest scores in the PedsQL CP module version 3.0 were observed in the Daily Activities ( $5.34 \pm 10.87$ ), and School Activities domains ( $8.15 \pm 18.65$ ). Sociodemographic and anthropometric data, including body mass index (BMI)-for-age, MUAC, body fat, triceps, and subscapular skinfold measurement, were predictors for the School Activities domain [ $F(11, 44) = 3.981$ ,  $P < 0.005$ ,  $R^2 = 0.499$ ]. Conclusions: Poor nutritional status in individuals with severe CP has been shown to negatively impact caregivers' quality of life. Therefore, a multidisciplinary approach involving nutritional intervention is essential to improve dietary provision and the nutritional status of children with CP.

PMID: [37814717](#)

### 16. Bone alterations of pamidronate therapy in children with cerebral palsy complicating orthopedic management

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J Pediatr Orthop B. 2023 Oct 9. doi: 10.1097/BPB.0000000000001136. Online ahead of print.

Cerebral palsy (CP) is a heterogeneous group of disorders with different clinical types and underlying genetic variants. Children with CP are at risk for fragility fractures secondary to low bone mineral density, and although bisphosphonates are prescribed for the treatment of children with bone fragility, there is limited information on long-term bone impact and safety. Children with CP usually present overinflated bones, and the thickening of cortical bone by pamidronate treatment can potentially further narrow the medullary canal. Our purpose was to report bone alterations attributable to pamidronate therapy that impact orthopedic care in children with CP. The study consisted of 41 children with CP treated with pamidronate for low bone mineral density from 2006 to 2020. Six children presented unique bone deformities and unusual radiologic features attributed to pamidronate treatment, which affected their orthopedic care. The cases included narrowing of the medullary canal and sclerotic bone, atypical femoral fracture, and heterotopic ossification. Treatment with bisphosphonate reduced the number of fractures from 101 in the pretreatment period to seven in the post-treatment period ( $P < 0.001$ ). In conclusion, children with CP treated with bisphosphonate have a reduction in low-energy fractures; however, some fractures still happen, and pamidronate treatment can lead to bone alterations including medullary canal narrowing with sclerotic bone and atypical femoral fractures. In very young children, failure to remodel may lead to thin, large femoral shafts with cystic medullary canals. More widespread use of bisphosphonates in children with CP may make these bone alterations more frequent. Level of evidence: Level IV: Case series with post-test outcomes.

PMID: [37811568](#)

### 17. A - 111 Neuropsychological Performance in CDK-13 Related Disorder: a Multi-Year Case Study

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Arch Clin Neuropsychol. 2023 Oct 8:acad067.128. doi: 10.1093/arclin/acad067.128. Online ahead of print.

**Objective:** CDK13-related disorder (CDK13-RD) is a rare genetic condition first described in 2016 as involving congenital heart defects, dysmorphic facial features, and intellectual developmental disorder (CHDFIDD). Few studies have reported clinical presentations. There is very minimal information about cognitive and neuropsychological outcomes; however, developmental delays and intellectual disability are common. This study presents longitudinal neuropsychological outcomes of one adolescent with CKD13-RD. **Method:** A female patient underwent five neuropsychological evaluations over eight years due to learning concerns. Pregnancy complications included recurrent maternal infections/treatment and fetal heart deceleration. The patient was born at full-term with average birthweight. Patient's history includes developmental delays, cerebral palsy, learning disabilities, anxiety, atrial septal defect, abnormal MRI and EEG, submucous cleft palate, velopharyngeal insufficiency, and craniofacial dysmorphisms. Genetic tests confirmed a likely pathogenetic variant in the CDK13-gene at 9-years-old. **Results:** Patient's intellectual functioning varied during the five evaluations, with heterogeneous performance across indices. Verbal comprehension was often a strength in the low average to average ranges. Visual perception, nonverbal reasoning, fine motor coordination/planning, attention, and aspects of executive functioning were consistently impaired. Memory across evaluations was variable but broadly average. Caregiver reports revealed increasing executive and adaptive functioning concerns and fluctuating mood/behavioral concerns across evaluations. **Conclusions:** Patient's overall intellectual functioning is generally stronger than expected compared to the few available studies. This case adds to the scarce literature on neuropsychological outcomes in children with CKD13-RD and helps elucidate developmental trends given these serial results. Additional research is needed to better understand neuropsychological presentations and long-term outcomes in children with CKD13-RD.

PMID: [37807238](#)