NEWS
Cerebral Palsy Research

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# Interventions and Management

**1. Influence of the corticospinal tract wiring pattern on sensorimotor functional connectivity and clinical correlates of upper limb function in unilateral cerebral palsy.**
Simon-Martinez C, Jaspers E, Alaerts K, Ortibus E, Balsters J, Mailleux L, Blommaert J, Sleurs C, Klingels K, Amant F, Uyttebroeck A, Wenderoth N, Feys H.

Sci Rep. 2019 Jun 3;9(1):8230. doi: 10.1038/s41598-019-44728-9.

In children with unilateral cerebral palsy (uCP), the corticospinal tract (CST)-wiring patterns may differ (contralateral, ipsilateral or bilateral), partially determining motor deficits. However, the impact of such CST-wiring on functional connectivity remains unknown. Here, we explored resting-state sensorimotor functional connectivity in 26 uCP with periventricular white matter lesions (mean age (standard deviation): 12.87 m (±4.5), CST wiring: 9 contralateral, 9 ipsilateral, 6 bilateral) compared to 60 healthy controls (mean age (standard deviation): 14.54 (±4.8)), and between CST-wiring patterns. Functional connectivity from each M1 to three bilateral sensorimotor regions of interest (primary sensory cortex, dorsal and ventral premotor cortex) and the supplementary motor area was compared between groups (controls vs. uCP; and controls vs. each CST-wiring group). Seed-to-voxel analyses from bilateral M1 were compared between groups. Additionally, relations with upper limb motor deficits were explored. Aberrant sensorimotor functional connectivity seemed to be CST-dependent rather than specific from all the uCP population: in the dominant hemisphere, the contralateral CST group showed increased connectivity between M1 and premotor cortices, whereas the bilateral CST group showed higher connectivity between M1 and somatosensory association areas. These results suggest that functional connectivity of the sensorimotor network is CST-wiring-dependent, although the impact on upper limb function remains unclear.

PMID: 31160679

**2. Natural History of Spastic Hip Disease.**
Lins LAB, Watkins CJ, Shore BJ.

J Pediatr Orthop. 2019 Jul;39(Issue 6, Supplement 1 Suppl 1):S33-S37. doi: 10.1097/BPO.0000000000001347.

BACKGROUND: Hip displacement in children with cerebral palsy is common and related to a child's gross motor function. Progressive lateral hip displacement can result in severe pain, impaired function and quality of life. The purpose of this paper is to review the literature to identify the natural history of untreated spastic hip displacement in children with cerebral palsy. METHODS: A search of the literature was carried out using PubMed to identify papers describing the natural history of spastic hip displacement in children with cerebral palsy. Population-based studies, large retrospective cohort series, and randomized trials were included when available; expert opinion and case series were excluded. RESULTS: A total of 79 articles were reviewed. Articles were then subdivided into 4 main categories: epidemiology, pathophysiology, outcome, and prevention. The prevalence of hip displacement as defined as a migration percentage >30% was found to be 33% in pooled

population-based studies. The risk of hip displacement was found to be linked to a child's gross motor function. Hip surveillance programs have been shown to be sustainable and capable of preventing hip dislocation. The majority of children with hip dislocation do develop pain, decreased function, and impaired health-related quality of life (HRQOL). CONCLUSIONS: Children with cerebral palsy are at risk of progressive lateral hip displacement proportional to their gross motor function. Untreated progressive lateral hip displacement has been shown to negatively impact a child's HRQOL and hip surveillance can decrease the incidence of hip dislocation.

PMID: 31169645

**3. Improved motor function in a pre-ambulatory child with spastic bilateral cerebral palsy, using a custom rigid ankle-foot orthosis-footwear combination: A case report.**
Young J, Jackson S.

Prosthet Orthot Int. 2019 Jun 4:309364619852239. doi: 10.1177/0309364619852239. [Epub ahead of print]

BACKGROUND: Ankle-foot orthoses may be used in pre-ambulatory children with cerebral palsy; however, their effect on the acquisition of walking is unknown. This case report aims to evaluate the effect of an ankle-foot orthosis-footwear combination on the acquisition of walking in a single subject with cerebral palsy. CASE DESCRIPTION AND METHODS: This study reports the orthotic management of a single child with spastic bilateral cerebral palsy over a 15-month period, during which time the ability to independently stand and walk was acquired. Custom rigid ankle-foot orthoses were prescribed. Gait speed and Edinburgh Visual Gait Score were assessed with and without the orthoses. FINDINGS AND OUTCOMES: The subject developed the ability to stand and walk using an ankle-foot orthosis-footwear combination with a walker frame, and to a limited extent without a walker frame. The subject remained unable to take independent steps unless wearing the ankle-foot orthosis-footwear combination. Clinically significant differences in gait speed and Edinburgh Visual Gait Score were observed. CONCLUSION: An ankle-foot orthosis-footwear combination may aid the development of independent walking in some children with cerebral palsy. Further research on the effects of orthoses on the acquisition of walking ability in children with cerebral palsy is needed. CLINICAL RELEVANCE: Custom rigid ankle-foot orthoses combined with footwear may aid the development of independent standing and walking in some children with bilateral spastic cerebral palsy. This intervention may be considered in clinical practice and future research in this patient group.

PMID: 31165679

**4. A commentary on Kalkman et al.'s letter to the editor regarding Alexander et al. (2019): "Children with cerebral palsy have larger in-vivo and linearly scaled Achilles tendon moment arms than typically developing children".**
Donnelly CJ, Alexander CF, Stannage K, Reid S.

J Biomech. 2019 May 9. pii: S0021-9290(19)30324-0. doi: 10.1016/j.jbiomech.2019.04.047. [Epub ahead of print]

PMID: 31171371

**5. Letter to the editor regarding: Effects of cerebral palsy on Achilles tendon moment arm length - Do children with CP have larger or smaller moment arms than typically developing children? Commentary on: Alexander et al.**
Kalkman BM, Maganaris CN, Bar-On L, O'Brien TD.

J Biomech. 2019 May 9. pii: S0021-9290(19)30323-9. doi: 10.1016/j.jbiomech.2019.03.039. [Epub ahead of print]

PMID: 31164223

**6. Effects of Radial Shockwave Therapy and Orthotics Applied with Physical Training on Motor Function of Children with Spastic Diplegia: A Randomized Trial.**
Elnaggar RK, Abd-Elmonem AM.

Phys Occup Ther Pediatr. 2019 May 31:1-16. doi: 10.1080/01942638.2019.1597821. [Epub ahead of print]

Aims: To explore the effects of radial shockwave therapy (rSWT) combined with standard orthotic management (SOM) on spasticity, functional balance, and gait in children with spastic diplegia. Methods: Sixty children with diplegia were allocated to group I (rSWT, n = 20), group II (SOM, n = 20), or group III (rSWT + SOM, n = 20). All groups received a physical training program 3 times/week for 3 months. Assessments were completed before and immediately after the intervention and included the Hoffman reflex/Myogenic response ratio of the soleus muscle (H/M ratio), balance, and gait. Results: At a significance criterion adjusted to $p < .006$, there were no between-group differences in balance or gait ($p > .006$). The rSWT + SOM group had a greater improvement of H/M ratio compared to rSWT alone ($p = .001$) but not to SOM alone ($p = .04$). Within-group analysis demonstrated significant improvement of all variables for rSWT + SOM ($p < .006$). The H/M ratio and knee midstance angle exhibited clinically meaningful improvement for rSWT alone ($p < .006$). No significant changes were observed in any variable for SOM alone ($p > .006$). Conclusions: Radial shockwave and orthotics together, or either of them along with physical training did not differ in improving balance or gait. Their combination was more effective than rSWT alone in reducing spasticity.

PMID: 31148494

**7. Predictors of Walking Efficiency in Children With Cerebral Palsy: Lower-Body Joint Angles, Moments, and Power.**
Noorkoiv M, Lavelle G, Theis N, Korff T, Kilbride C, Baltzopoulos V, Shortland A, Levin W, Ryan JM.

Phys Ther. 2019 Jun 1;99(6):711-720. doi: 10.1093/ptj/pzz041.

BACKGROUND: People with cerebral palsy (CP) experience increased muscle stiffness, muscle weakness, and reduced joint range of motion. This can lead to an abnormal pattern of gait, which can increase the energy cost of walking and contribute to reduced participation in physical activity. OBJECTIVE: The aim of the study was to examine associations between lower-body joint angles, moments, power, and walking efficiency in adolescents with CP. DESIGN: This was a cross-sectional study. METHODS: Sixty-four adolescents aged 10 to 19 years with CP were recruited. Walking efficiency was measured as the net nondimensional oxygen cost (NNcost) during 6 minutes of overground walking at self-selected speed. Lower-body kinematics and kinetics during walking were collected with 3-dimensional motion analysis, synchronized with a treadmill with integrated force plates. The associations between the kinematics, kinetics, and NNcost were examined with multivariable linear regression. RESULTS: After adjusting for age, sex, and Gross Motor Function Classification System level, maximum knee extension angle ($\beta = -0.006$), hip angle at midstance ($\beta = -0.007$), and maximum hip extension ($\beta = -0.008$) were associated with NNcost. Age was a significant modifier of the association between the NNcost and a number of kinematic variables. LIMITATIONS: This study examined kinetic and kinematic variables in the sagittal plane only. A high interindividual variation in gait pattern could have influenced the results. CONCLUSIONS: Reduced knee and hip joint extension are associated with gait inefficiency in adolescents with CP. Age is a significant factor influencing associations between ankle, knee, and hip joint kinematics and gait efficiency. Therapeutic interventions should investigate ways to increase knee and hip joint extension in adolescents with CP.

PMID: 31155663

**8. Real-Time Detection of Seven Phases of Gait in Children with Cerebral Palsy Using Two Gyroscopes.**
Behboodi A, Zahradka N, Wright H, Alesi J, Lee SCK.

Sensors (Basel). 2019 Jun 1;19(11). pii: E2517. doi: 10.3390/s19112517.

A recently designed gait phase detection (GPD) system, with the ability to detect all seven phases of gait in healthy adults, was modified for GPD in children with cerebral palsy (CP). A shank-attached gyroscope sent angular velocity to a rule-based algorithm in LabVIEW to identify the distinct characteristics of the signal. Seven typically developing children (TD) and five children with CP were asked to walk on treadmill at their self-selected speed while using this system. Using only shank angular velocity, all seven phases of gait (Loading Response, Mid-Stance, Terminal Stance, Pre-Swing, Initial Swing, Mid-Swing and Terminal Swing) were reliably detected in real time. System performance was validated against two established GPD methods: (1) force-sensing resistors (GPD-FSR) (for typically developing children) and (2) motion capture (GPD-MoCap) (for both typically developing children and children with CP). The system detected over 99% of the phases identified by GPD-FSR and GPD-MoCap. Absolute values of average gait phase onset detection deviations relative to GPD-MoCap were less than 100 ms for both TD children and children with CP. The newly designed system, with minimized sensor setup and low processing burden, is cosmetic and economical, making it a viable solution for real-time stand-alone and portable applications such as triggering functional electrical stimulation (FES) in rehabilitation systems. This paper verifies the applicability of the GPD system to identify specific gait events for triggering FES to enhance gait in children with CP.

PMID: 31159379

**9. Exploring Physiotherapists' Use of Motor Learning Strategies in Gait-Based Interventions for Children with Cerebral Palsy.**
Ryan JL, Wright FV, Levac DE.

Phys Occup Ther Pediatr. 2019 Jun 3:1-14. doi: 10.1080/01942638.2019.1622623. [Epub ahead of print]

Aim: This study investigated physiotherapists' experiences using motor learning strategies (MLS) in gait-based interventions for children with cerebral palsy (CP). The objectives were to explore how child characteristics, physiotherapist decision-making, and treatment approach influenced intentional MLS use. Methods: Semi-structured interviews were conducted with eight physiotherapists who provided gym- and/or Lokomat-based treatment to children with CP. Interviews were analyzed using directed content analysis and a modified constant comparison method. Results: Three themes described their experiences: (1) MLS use is driven by the unique aspects of the child, physiotherapist, and intervention; (2) The use and description of motor learning content varies among physiotherapists; and (3) The Lokomat is "the same but different." Child characteristics were at the forefront of MLS selection in both interventions. The terminology used to describe MLS use varied considerably among therapists. They used similar clinical decision-making in gym- and Lokomat-based interventions. Conclusions: Conscious reflection on the factors affecting MLS use could facilitate related clinical decision-making in physiotherapy interventions for children with CP. Increased awareness of MLS and use of a structured framework for reporting MLS are required to promote intentional MLS use and generate CP-specific evidence-based MLS research.

PMID: 31154883

**10. The impact of centre of pressure error on predicted joint kinetics during cerebral palsy and typically developed gait: A clinical perspective.**
Brady K, Kiernan D.

J Biomech. 2019 May 24. pii: S0021-9290(19)30376-8. doi: 10.1016/j.jbiomech.2019.05.034. [Epub ahead of print]

Centre of Pressure (CoP) location error is common when using kinematic and kinetic data to predict intersegmental forces and net joint moments during gait. Changes in peak moments due to CoP error have been reported in the literature. However, debate exists as to what levels of error are acceptable. The aim of this study was to examine the impact of CoP error on the kinetic profiles of children with typical development (TD) and children with cerebral palsy (CP) during gait. Three-dimensional kinematic and kinetic data were recorded and simulated CoP errors were applied at 3 mm, 6 mm, 9 mm, 12 mm increments in both positive and negative anteroposterior and mediolateral directions. Absolute differences in maximum kinetic parameters between increments were assessed in conjunction with changes in the Gait Deviation Index-Kinetic (GDI-Kinetic). Changes in GDI-Kinetic above 3.6 points were considered clinically significant. Maximum peak changes of up to 24.8% (CP) and 34.7% (TD) (sagittal plane) and up to 36.8% (CP) and 61.5% (TD) (coronal plane) were demonstrated at the knee. While absolute percentage differences were high at some error increments, GDI-Kinetic results suggested that such large percentage differences may still be clinically acceptable. Children with TD demonstrated clinically significant changes in GDI-Kinetic for CoP displacements of 9 mm and 12 mm, corresponding to 23% and 35% absolute differences in maximum moments. In contrast, the clinically significant threshold was not reached for children with CP that may be related to a slower walking speed. The findings of this study highlight the need for laboratories to consider the thresholds currently used for CoP error, which will help guide quality assurance procedures.

PMID: 31153622

**11. Use of a Novel Functional Electrical Stimulation Gait Training System in 2 Adolescents With Cerebral Palsy: A Case Series Exploring Neurotherapeutic Changes.**
Behboodi A, Zahradka N, Alesi J, Wright H, Lee SCK.

Phys Ther. 2019 Jun 1;99(6):739-747. doi: 10.1093/ptj/pzz040.

BACKGROUND AND PURPOSE: Cerebral palsy (CP) is characterized by decreased passive joint range-of-motion and impaired walking, resulting in progressive loss of function. Typical gait training interventions for children with CP appear insufficient to mitigate these effects. The purpose of this case report is to describe the use of a new treadmill-based gait training intervention using active correction with functional electrical stimulation (FES) in 2 adolescents with CP. CASE DESCRIPTION: Two participants with CP (13-year-old girls, Gross Motor Function Classification System [GMFCS] level II and III) trained by walking on a treadmill, with FES assistance, for 30 minutes, 3 times per week, for 12 weeks. The

intervention used a feedback control system to detect all 7 phases of gait in real time and triggered FES to the appropriate muscle groups (up to 5 bilaterally) based on the detected gait phase. Joint kinematics, step width, stride length, walking endurance, peak oxygen uptake ($\dot{v}^{o}_{2}$), and oxygen (O2) cost of walking were evaluated preintervention and postintervention. OUTCOMES: Both participants showed improved knee and ankle angles and step width relative to children who are typically developing, and both exhibited increased stride length. One participant (GMFCS III) improved peak $\dot{v}^{o}_{2}$and walking endurance but not O2 cost of walking at her original self-selected walking speed. The other participant (GMFCS II) improved O2 cost of walking but not peak $\dot{v}^{o}_{2}$ or walking endurance. These differences are partly explained by differences in gait type, functional abilities, and initial fitness levels. Most improvements persisted at follow-up, indicating short-term neurotherapeutic effects. DISCUSSION: Most improvements persisted at follow-up, suggesting short-term neurotherapeutic effects. This case series demonstrates the promising utility of FES-assisted gait-training interventions, tailored to target individual gait deviations, in improving walking performance.

PMID: 31155665

**12. The effects of electromyography-assisted modelling in estimating musculotendon forces during gait in children with cerebral palsy.**
Veerkamp K, Schallig W, Harlaar J, Pizzolato C, Carty CP, Lloyd DG, van der Krogt MM.

J Biomech. 2019 May 22. pii: S0021-9290(19)30358-6. doi: 10.1016/j.jbiomech.2019.05.026. [Epub ahead of print]

Neuro-musculoskeletal modelling can provide insight into the aberrant muscle function during walking in those suffering cerebral palsy (CP). However, such modelling employs optimization to estimate muscle activation that may not account for disturbed motor control and muscle weakness in CP. This study evaluated different forms of neuro-musculoskeletal model personalization and optimization to estimate musculotendon forces during gait of nine children with CP (GMFCS I-II) and nine typically developing (TD) children. Data collection included 3D-kinematics, ground reaction forces, and electromyography (EMG) of eight lower limb muscles. Four different optimization methods estimated muscle activation and musculotendon forces of a scaled-generic musculoskeletal model for each child walking, i.e. (i) static optimization that minimized summed-excitation squared; (ii) static optimization with maximum isometric muscle forces scaled to body mass; (iii) an EMG-assisted approach using optimization to minimize summed-excitation squared while reducing tracking errors of experimental EMG-linear envelopes and joint moments; and (iv) EMG-assisted with musculotendon model parameters first personalized by calibration. Both static optimization approaches showed a relatively low model performance compared to EMG envelopes. EMG-assisted approaches performed much better, especially in CP, with only a minor mismatch in joint moments. Calibration did not affect model performance significantly, however it did affect musculotendon forces, especially in CP. A model more consistent with experimental measures is more likely to yield more physiologically representative results. Therefore, this study highlights the importance of calibrated EMG-assisted modelling when estimating musculotendon forces in TD children and even more so in children with CP.

PMID: 31153626

**13. Three by three weeks of robot-enhanced repetitive gait therapy within a global rehabilitation plan improves gross motor development in children with cerebral palsy - a retrospective cohort study.**
Weinberger R, Warken B, König H, Vill K, Gerstl L, Borggraefe I, Heinen F, von Kries R, Schroeder AS.

Eur J Paediatr Neurol. 2019 May 18. pii: S1090-3798(18)30423-9. doi: 10.1016/j.ejpn.2019.05.003. [Epub ahead of print]

AIM: To assess the improvement in gross motor function following three blocks of a three-week, intensive robot-enhanced treadmill therapy (ROBERT-Program). METHOD: retrospective chart review in a before-after interventional trial in children with cerebral palsy attending a university hospital outpatient rehabilitation centre. Patients received three blocks of a three-week, 12 sessions ROBERT-Program over a mean period of 24 months. Outcome measures were block specific and cumulative improvement in GMFM 66, D and E. Longterm GMFM 66 improvements were compared to the individuals' expected increment as derived from previously published GMFM-66 percentiles. 95% confidence intervals (CI) and paired t-test were calculated. RESULTS: 20 children (8 GMFCS Level II; 12 GMFCS Level III, mean age 5.9 years (CI: [5.0; 6.7])) were treated. For each block a significant increase in motor performance in similar size could be observed without deterioration between blocks. The cumulative improvement during 21 months observation period was: 6.5 (CI: [4.8; 8.2]) in GMFM 66, which represents a clinically meaningful effect size of 3.6 (CI: [1.4; 5.8]) above the expected improvement. INTERPRETATION: Progressive clinically meaningful improvement in motor performance for three blocks of ROBERT-Program was observed. Cumulative GMFM 66 improvements exceeded the individuals' age-specific expected course.

PMID: 31155454

**14. Robot Reinforcement and Error-Based Movement Learning in Infants With and Without Cerebral Palsy.**
Kolobe THA, Fagg AH.

Phys Ther. 2019 Jun 1;99(6):677-688. doi: 10.1093/ptj/pzz043.

BACKGROUND: Prone mobility, central to development of diverse psychological and social processes that have lasting effects on life participation, is seldom attained by infants with cerebral palsy (CP) and has no tested interventions. Reinforcement learning (RL) and error-based movement learning (EBL) offer novel intervention possibilities. OBJECTIVE: This study examined movement learning strategies in infants with or at risk for CP using RL and EBL during acquisition of prone locomotion. DESIGN: The study was a randomized trial that used repeated measures. SETTING: The study setting was a university physical therapy clinic in the United States. PATIENTS: Thirty infants aged 4.5 to 6.5 months participated in the study: 24 had or were at risk for CP, and 6 were typically developing. INTERVENTION: Infants with and at risk for CP were randomly assigned to a combination of RL and EBL (SIPPC-RE), or RL only (SIPPC-R) conditions. Infants with typical development comprised the RL-only reference group (SIPPC-TD). Infants trained in prone locomotion with the Self-Initiated Prone Progression Crawler (SIPPC) robotic system for three 5-minute trials, twice a week for 12 weeks in their homes or child care. All training sessions were videotaped for behavioral coding. MEASUREMENTS: The SIPPC gathered robot and infant trunk/limb movement data. Randomized 2-way analysis of variance with repeated measures and Pearson r to analyze the data was used. RESULTS: Results included the number of arm movements and trial-and-error activity distinguished between the SIPPC-RE and SIPPC-R groups. The mean change in arm movements from baseline for the SIPPC-RE and SIPPC-R groups was 4.8 m and -7.0 m, respectively. The mean differences in rotational amplitude (trial and error) from baseline to the end of the study were 278 degrees and 501 degrees, respectively. These changes were correlated with distance traveled and goal-directed movements. The latter increased over the 12 weeks for the SIPPC-RE and SIPPC-TD groups, but not the SIPPC-R group. LIMITATIONS: The CP groups were unequal due to reassignment and did not include a typically developing comparison group of a combination of RL and EBL. CONCLUSION: These findings suggest movement learning and retention in infants with CP is differentially affected by the use of RL and EBL, with a combination of both showing more promise than RL alone. The findings also implicate cognition, type of brain insult, emergence of reaching, and muscle force production, which must be explored in future studies.

PMID: 31155667

**15. Cross-Cultural Validation Study of the Japanese Version of the ABILOCO-Kids in Ambulatory Children With Cerebral Palsy Using Rasch Analysis.**
Himuro N, Nishibu H, Abe H, Mori M.

Phys Occup Ther Pediatr. 2019 Jun 4:1-13. doi: 10.1080/01942638.2019.1609150. [Epub ahead of print]

Aims: To determine the structural validity, construct validity, cross-cultural validity, internal consistency and test-retest reliability of the Japanese version of the ABILOCO-Kids in children with cerebral palsy. Methods: One-hundred sixteen parents of children with cerebral palsy (Gross Motor Function Classification System level I [n = 66], II [n = 32], and III [n = 18]) reported on walking ability using the Japanese version of the ABILOCO-Kids. For test-retest reliability, 23 participants were evaluated. Results: The mean ABILOCO-Kids logit score was 2.48 (range -7.44 to 5.83). Rasch analysis and principal component analysis were used to determine the structural validity. The construct validity was confirmed on the basis of differences in the ABILOCO-Kids scores among the Gross Motor Function Classification System levels. Cronbach's α and the item-to-total correlation coefficient supported the internal consistency. The intra-class correlation coefficient was 0.96, standard error of measurement 0.56 and minimal detectable change 1.55. The cross-cultural validity analyses showed differential item functioning according to the analyses of the invariance of item difficulty and person's ability estimates. Conclusions: The ABILOCO-Kids is a reliable and valid measure of walking ability in children with cerebral palsy in Japan. The lack of evidence on cross-cultural validity indicates we should interpret results cautiously in cases of international comparison.

PMID: 31164026

**16. Use of outcome measures in children with severe cerebral palsy: A survey of U.K. physiotherapists.**
Knox V, Vuoskoski P, Mandy A.

Physiother Res Int. 2019 Jun 6:e1786. doi: 10.1002/pri.1786. [Epub ahead of print]

OBJECTIVES: To investigate the use of outcome measures for children with cerebral palsy (CP) by paediatric physiotherapists (PTs) who are based in the United Kingdom, as limited research exists regarding their use in this population, and to explore therapists' use of measures within different Gross Motor Function Classification System (GMFCS) levels and for different types of CP. METHODS: A six-item online survey was advertised through two paediatric therapy special interest groups inviting physiotherapists to participate. Descriptive statistics (range, frequencies, and percentages) were used to analyse survey data. RESULTS: Two hundred and seven physiotherapists returned completed survey questionnaires. One hundred and seventy -six (85%) therapists reported using a wide variety of outcome measures (57). Eighty-one per cent of therapists' responses for having "most difficulty" in identifying responsive outcome measures were for children with impaired mobility at GMFCS Levels IV-V and children with more significant body involvement (76%), for example, spastic quadriplegia or dyskinesia. Eighty-six per cent of therapists' responses for having "greatest ease" of identifying responsive outcome measures were for children within GMFCS Levels I-III and for children with less significant body involvement (72%), for example, hemiplegia. The variety of outcome measures used by therapists with children within GMFCS IV-V was less (16). CONCLUSIONS: The majority (85%) of the PTs surveyed used outcome measures with children with CP, but 81% perceived a difficulty in identifying responsive measures for children with more severe impairments who are classified as GMFCS IV-V. The reasons for this perceived difficulty warrant investigation and may suggest a need for training regarding relevant measures and/or a need to develop new measures for this group of children.

PMID: 31172663

**17. Children and Youth with Complex Cerebral Palsy: Care and Management.**
Kuenzle C.

Neuropediatrics. 2019 Jun 3. doi: 10.1055/s-0039-1685523. [Epub ahead of print]

PMID: 31158910

**18. Criterion validity of assessment methods to estimate body composition in children with cerebral palsy: A systematic review.**
Snik DAC, de Roos NM.

Ann Phys Rehabil Med. 2019 May 31. pii: S1877-0657(19)30069-7. doi: 10.1016/j.rehab.2019.05.003. [Epub ahead of print]

BACKGROUND: Poor nutritional status is a problem in a high number of children with cerebral palsy (CP) and impairs their well-being. Therefore, periodic assessment of nutritional status and especially body composition is essential. However, we lack consensus on the best method to assess body composition in clinical practice. OBJECTIVE: We aimed to systematically review the available evidence on the criterion validity of equation-based skinfold measurement and bioelectrical impedance analysis (BIA) to estimate body composition in children with CP. METHODS: In a systematic review (MEDLINE, Cochrane Library and EMBASE), we identified studies that reported on the agreement between the estimation of body composition by equations of skinfold thickness or impedance values of BIA with a gold standard (isotope techniques or dual-energy-X-ray-absorptiometry [DXA]) in children with CP. We included only studies that provided correlations or agreement between estimations of body compartments (e.g., percentage body fat [%BF] or fat mass). Limits of agreement of 2.5%BF points were considered acceptable. Study quality was assessed by using the Quality Assessment of Diagnostic Accuracy Studies 2. RESULTS: We included reports of 9 studies describing 3 skinfold equations and 4 equations to estimate body composition with BIA. Neither skinfold equations nor BIA could be reliably used to assess body composition in an individual child with CP at one point in time. On a population level, the Gurka skinfold equation was valid in ambulant children with CP, and the Kushner and Fjeld BIA equations were valid in a heterogeneous group of children with CP. Conclusions The future role of skinfold equations and BIA to assess and monitor body composition in an individual child with CP needs to be further investigated.

PMID: 31158551

**19. Quantitative analysis of vowel production in cerebral palsy children with dysarthria.**
Mou Z, Teng W, Ouyang H, Chen Y, Liu Y, Jiang C, Zhang J, Chen Z.

J Clin Neurosci. 2019 Jun 1. pii: S0967-5868(19)30603-4. doi: 10.1016/j.jocn.2019.05.020. [Epub ahead of print]

OBJECTIVE: The present study aimed to identify certain acoustic parameters for speech evaluation in cerebral palsy children with dysarthria. METHODS: The subject included 30 native Mandarin-Speaking children with cerebral palsy, who were 5-15 years old, and 13 healthy children in a similar age range. Each subject was recorded while producing a list of 12 Mandarin words, which included three syllables ('ba', 'bi' and 'du'), in all four Mandarin tones. The formants (F1 and F2) of monophthong vowels /a, i, u/ were extracted from each vowel token. Based on F1 and F2, the vowel acoustic indexes VSA, VAI and FCR were calculated and analyzed. RESULTS: Compared with the control group, the cerebral palsy group had significantly low F1 and F2 in vowel /a/ ($P < 0.05$), and F2 in vowel /i/ ($P < 0.05$), while F1 and F2 in vowel /u/ and F1 in vowel /i/ had no significant difference. Between the healthy group and cerebral palsy group, the differences in VSA, VAI and FCR were all statistically significant. CONCLUSION: Children with cerebral palsy have reduced vowel space and speech articulation. The significant difference in vowel acoustic indexes (VSA, VAI and FCR) among the two groups revealed that the three indexes were sensitive to the variation of the vowels production in children with cerebral palsy, and that these may be used as an evaluation method of speech intelligibility caused by impaired vowel pronunciation in children with cerebral palsy, and the effect of rehabilitation therapy.

PMID: 31164264

**20. Laryngotracheal separation in pediatric patients: 13-year experience in a reference service.**
Antunes LA, Talini C, Carvalho BCN, Guerra JP, Aristides EDS, Oliveira DE, Avilla SGA.

Einstein (Sao Paulo). 2019 Jun 3;17(3):eAO4467. doi: 10.31744/einstein_journal/2019AO4467. [Article in English, Portuguese]

OBJECTIVE: To evaluate clinical stability of neurologically impaired children and adolescents with recurrent pneumonia submitted to laryngotracheal separation. METHODS: Between October 2002 and June 2015, 92 neurologically impaired children from a reference service, with median age of 68.5 months were submitted to laryngotracheal separation. Data were evaluated and statistical analysis was made by Student's t test and Pearson's $\chi 2$ test (significance level adopted of 95%). RESULTS: Fifty-three children were male (57.6%). Forty-six children required admission to intensive care, and 42.4% needed mechanical ventilation. We observed that 90.2% of patients were exclusively fed by gastrostomy and 72.4% of the gastrostomies were performed before the tracheal surgery. Thirteen (14.1%) children had postoperative complications as follows: fistulae (5.4%), bleeding (4.3%), granuloma (2.2%) and stenosis (3.2%). A total of 24 patients had pneumonia in the postoperative period (26.1%), but there was a significant drop in occurrence of this condition after surgery (100% versus 26.1%; $p<0.001$). Twenty-three patients (25%) died. Postoperative complications were similar when comparing patients who died and those that presented good outcome (16.7% versus 13.2%; $p=0.73$). CONCLUSION: When well-indicated, the laryngotracheal separation reduces the incidence of postoperative pulmonary infections, thus improving quality of life and reducing admissions to hospital. Laryngotracheal separation should be indicated as a primary procedure in patients with cerebral palsy and recurrent aspiration pneumonia.

PMID: 31166409

**21. Randomised controlled trial of a novel online cognitive rehabilitation programme for children with cerebral palsy: a study protocol.**
Wotherspoon J, Whittingham K, Boyd RN, Sheffield J.

BMJ Open. 2019 Jun 4;9(6):e028505. doi: 10.1136/bmjopen-2018-028505.

INTRODUCTION: Cerebral palsy (CP) is the most common cause of physical disability in children, with an estimated 600-700 infants born with CP in Australia each year. CP is typically associated with motor impairments, but nearly half of all children with CP also experience cognitive impairment, potentially impacting educational and vocational achievement. This paper reports the protocol for a randomised controlled trial of a computerised cognitive training intervention based on behavioural principles: Strengthening Mental Abilities through Relational Training (SMART). The study aims to investigate SMART's effect on fluid reasoning, executive function and academic achievement in children with CP. METHODS AND ANALYSIS: Sixty children with mild to moderate CP (Gross Motor Function Classification Scale I-IV) aged between 8 years and 12 years will be recruited. Participants will be randomly allocated to two groups: SMART cognitive training and waitlist control. Families will access the programme at home over a 4-month period. Assessments will be administered at baseline, 20 weeks and at 40 week follow-up for retention. The primary outcome will be fluid intelligence, while academic achievement, executive function and social and emotional well-being will be secondary outcomes. ETHICS AND DISSEMINATION: This study has approval from the Children's Health Queensland Hospital and Health Service Research Ethics Committee (HREC/14/QRCH/377) and The University of Queensland (2017001806). If the computerised cognitive training programme is found to be effective, dissemination of these findings would assist children with CP by providing an accessible, cost-effective intervention

that can be completed at home at the individual's own pace. REGISTRATION DETAILS: The study was registered prospectively on 10 November 2017 to present. Recruitment is now under way, and we aim to complete recruitment by June 2019, with data collection finalised by March 2020. TRIAL REGISTRATION NUMBER: ACTRN12617001550392; Pre-results.

PMID: 31167872

**22. Content validation of the Quality of Life Inventory - Disability (QI-Disability).**
Epstein A, Williams K, Reddihough D, Murphy N, Leonard H, Whitehouse A, Jacoby P, Downs J.

Child Care Health Dev. 2019 Jun 4. doi: 10.1111/cch.12691. [Epub ahead of print]

BACKGROUND: Focus is shifting to better understand the lived experiences of children with intellectual disability in relation to their quality of life (QOL). Yet no available QOL measures are grounded in the domains important for this population. We previously conducted qualitative parent caregiver interviews identifying QOL domains in children with intellectual disability to constitute a new measure of QOL. This study describes the content validity of the Quality of Life Inventory - Disability (QI-Disability), a parent-report measure developed for children with intellectual disability. METHODS AND RESULTS: Questionnaire items were extracted from a qualitative dataset of 77 parent caregiver interviews. To establish content validation, a draft of QI-Disability was administered to 16 parent caregivers of children with intellectual disability (Down syndrome, Rett syndrome, cerebral palsy, or autism spectrum disorder). Parents participated in a cognitive interviewing procedure known as the "think-aloud" method. The process of item generation, cognitive debriefing and refinement of QI-Disability prior to its pilot testing are described. A conceptual framework is presented. CONCLUSIONS: Satisfactory content validity is reported, where ongoing consumer feedback shaped the dataset from which the final items were selected. Use of QI-Disability for children with intellectual disability will allow for greater insight into service utility and targeted intervention. 

PMID: 31163096

**23. Quality of life and burden of caregivers of children and adolescents with disabilities.**
Barros ALO, de Gutierrez GM, Barros AO, Santos MTBR.

Spec Care Dentist. 2019 Jun 6. doi: 10.1111/scd.12400. [Epub ahead of print]

AIMS: To evaluate the quality of life (QoL) and burden of primary caregivers of children and young adults (PCCYAs) with and without disabilities. METHODS: A cross-sectional study was carried out with sample composed of 336 PCCYAs with cerebral palsy (CP; n = 84), Down syndrome (DS; n = 84), autism spectrum disorder (ASD; n = 84), and without disabilities (control group: CG n = 84), matched by gender and age. The burden of caregivers was assessed with the Zarit Burden Interview (ZBI), whereas QoL was assessed using the WHOQOL-BREF instrument. RESULTS: QoL and burden of CG presented better results compared to groups with disabilities, with the lowest environmental domain of all study groups (P <.001). The prevalence of burden was moderate for PCCAs of groups with disabilities. There was association between all WHOQOL-BREF and ZBI domains and variables age, schooling, occupation and per capita income (Spearman's correlation coefficient, P <.05). There is a negative impact on WHOQOL-BREF, with an increase in the level of burden of PCCAs with disabilities. CONCLUSION: The majority of PCCYAs were unemployed married mothers, with low schooling and health problems. Older caregivers experience even higher burden and greater impact on QoL.

PMID: 31172548

**24. The quality of life of polish children with cerebral palsy and the impact of the disease on the family functioning.**
Kołtuniuk A, Rozensztrauch A, Budzińska P, Rosińczuk J.

J Pediatr Nurs. 2019 May 29. pii: S0882-5963(18)30493-7. doi: 10.1016/j.pedn.2019.05.011. [Epub ahead of print]

PURPOSE: Care and upbringing of a child with cerebral palsy (CP) may affect the functioning of parents and the whole family. This study aimed to evaluate the quality of life (QOL) of children with CP in parents' opinion and the impact of disease on family functioning. DESIGN AND METHODS: This cross-sectional study was conducted among 100 parents of children

with CP. Survey instruments used included an Authors-Designed Questionnaire (ADQ) to collect sociodemographic and educational background data as well as four standardized questionnaires for pediatric QOL (PedsQL-GC, PedsQL-CPM, PedsQL-FIM) and for life satisfaction (SWLS). RESULTS: Teenagers were characterised by a higher QOL compared to other age groups. The lowest scores were observed in the PedsQL-CPM domain of daily and school activities and in the physical functioning domain of the PedsQL. It was shown that family functioning is affected by the children's age and place of residence as well as the level of parental education. It was also shown that men, parents in a relationship, those living in the city and those with vocational education were characterised by a higher level of satisfaction with life than other groups. CONCLUSIONS: The QOL of children with CP is reduced compared to the QOL of healthy children and their condition has a significant impact on family functioning. Therefore, learning about the factors that influence QOL will allow health care providers to properly plan actions aimed at minimising the negative impact of CP on children's QOL and improving the functioning of their families.

PMID: 31153684

**25. Neurodevelopmental outcome at early school age in a Swiss national cohort of very preterm children.**
Pittet-Metrailler MP, Mürner-Lavanchy I, Adams M, Bickle-Graz M, Pfister RE, Natalucci G, Grunt S, Tolsa CB, Swiss National Network And Follow-Up Group.

Swiss Med Wkly. 2019 Jun 2;149:w20084. doi: 10.4414/smw.2019.20084. eCollection 2019 May 20.

BACKGROUND: Infants born very preterm are at higher risk of long-term neurodevelopmental problems than children born at term. Although there are increasing numbers of reports on outcomes from international cohorts of premature infants, a Swiss national report on infants after 2 years of age is lacking. AIMS OF THE STUDY: To describe neurodevelopmental outcomes at early school age of preterm children born in Switzerland with a special focus on the cognitive abilities. METHODS: This prospective national cohort study included children born alive before 30 weeks of gestation in 2006. At 5 years of age, children underwent a neurological examination and intelligence testing with the Kaufman Assessment Battery for Children first edition (K-ABC). We assessed the mental processing composite score (MPC) and its subscales to explore specific cognitive deficits. The primary outcome was cognitive impairment (MPC score <-1 standard deviation from the normative mean), motor impairment (cerebral palsy), or sensory impairment (any visual or hearing deficiency). The need for early intervention or therapies and the association of perinatal factors with cognitive impairment were secondary and tertiary outcomes. Logistic regression models were used to analyse associations between neonatal factors and cognitive outcome. RESULTS: Of 289 survivors, 235 were assessed. Of the 199 children with results obtained from the K-ABC, 42 (21%) showed cognitive impairment and 80 (40%) showed impairment in short-term memory. Cerebral palsy was diagnosed in 14 (6%), and visual and auditory impairment in 36 (15%) and 12 (5%) children, respectively; 63 (27%) needed early intervention or therapies. Cognitive impairment was associated with low socioeconomic status, but not with gestational age, small birthweight for gestational age, bronchodysplasia, or significant brain injury. A total of 146 children (63%) survived without any impairment. CONCLUSION: This is the first study to report neurodevelopmental outcomes at early school age in a Swiss cohort. The majority had favourable outcomes, but 21% of children demonstrated cognitive impairment, which was most pronounced in short-term memory. Our findings were similar to those of international cohorts and indicate that preterm children born before 300/7 gestational weeks, especially those living in unfavourable social environments, are at an increased risk of cognitive impairment and need close monitoring beyond early school age. Trial registration no: KEK-ZH-Nr.2014-0552  .

PMID: 31154661

**26. Mental fatigue level detection based on event related and visual evoked potentials features fusion in virtual indoor environment.**
Lamti HA, Ben Khelifa MM, Hugel V.

Cogn Neurodyn. 2019 Jun;13(3):271-285. doi: 10.1007/s11571-019-09523-2. Epub 2019 Jan 29.

The purpose of this work is to set up a model that can estimate the mental fatigue of users based on the fusion of relevant features extracted from Positive 300 (P300) and steady state visual evoked potentials (SSVEP) measured by electroencephalogram. To this end, an experimental protocol describes the induction of P300, SSVEP and mental workload (which leads to mental fatigue by varying time-on-task) in different scenarios where environmental artifacts are controlled (obstacles number, obstacles velocities, ambient luminosity). Ten subjects took part in the experiment (with two suffering from cerebral palsy). Their mission is to navigate along a corridor from a starting point A to a goal point B where specific flickering stimuli are introduced to perform the P300 task. On the other hand, SSVEP task is elicited thanks to 10 Hz flickering lights. Correlated features are considered as inputs to fusion block which estimates mental workload. In order to deal with uncertainties and heterogeneity of P300 and SSVEP features, Dempster-Shafer (D-S) evidential reasoning is introduced. As the

goal is to assess the reliability for the estimation of mental fatigue levels, D-S is compared to multi layer perception and linear discriminant analysis. The results show that D-S globally outperforms the other classifiers (although its performance significantly decreases between healthy and palsied groups). Finally we discuss the feasibility of such a fusion proposal in real life situation.

PMID: 31168331

**27. Survival and mortality in cerebral palsy: observations to the sixth decade from a data linkage study of a total population register and National Death Index.**
Blair E1,2, Langdon K3, McIntyre S4,5,6, Lawrence D7, Watson L8.

BMC Neurol. 2019 Jun 4;19(1):111. doi: 10.1186/s12883-019-1343-1.

BACKGROUND: Likely duration of survival of children described as having cerebral palsy is of considerable interest to individuals with cerebral palsy, their families, carers, health professionals, health economists and insurers. The aim of this paper is to describe patterns of survival and mortality to the sixth decade in a geographically defined population of people with cerebral palsy stratified according to the clinical description of their impairments in early childhood. METHODS: Identifiers of persons born in Western Australia 1956-2011, registered with cerebral palsy on the Western Australian Register of Developmental Anomalies and surviving at least 12 months, were linked to the Australian National Death Index in December 2014. Patterns of mortality were investigated using survival analysis methods. RESULTS: Of 3185 eligible persons, 436 (13.7%) had died. Of that sample the 22% with the mildest impairment had survival patterns similar to the general population. Mortality increased with increasing severity of impairment. Of 349 (75%) with available cause of death data, 58.6% were attributed to respiratory causes, including 171 (49%) to pneumonia at a mean age of 14.6 (sd 13.4) years of which 77 (45%) were attributed to aspiration. For the most severely impaired, early childhood mortality increased in succeeding decades of birth cohorts from 1950s to 1990 with 20% dying by 4 years of age in the 1981-1990 birth cohort; it then decreased for subsequent birth cohorts, 20% mortality not being attained until 15 years of age. However by 20 years of age mortality of the most severely impaired born in the 1991-2000 birth cohort exceeded that of all other birth cohorts. Remaining life expectancies by age to 50 years have been estimated for two strata with more severe impairments. CONCLUSION: For 22% of individuals with cerebral palsy with mild impairment survival to 58 years is similar to that of the general population. Since 1990 mortality for those with severe cerebral palsy in Western Australia has tended to shift from childhood to early adulthood.

PMID: 31164086

**28. Ascending Lipopolysaccharide-Induced Intrauterine Inflammation in Near-Term Rabbits Leading to Newborn Neurobehavioral Deficits.**
Shi Z, Vasquez-Vivar J, Luo K, Yan Y, Northington F, Mehrmohammadi M, Tan S.

Dev Neurosci. 2019 Jun 4:1-13. doi: 10.1159/000499960. [Epub ahead of print]

BACKGROUND: Chorioamnionitis from ascending bacterial infection through the endocervix is a potential risk factor for cerebral palsy. Tetrahydrobiopterin, an essential cofactor for nitric oxide synthase (NOS) and amino acid hydroxylases, when augmented in the fetal brain, prevents some of the cerebral palsy-like deficits in a rabbit hypoxia-ischemia model. OBJECTIVES: To study the effect of lipopolysaccharide (LPS)-induced intrauterine inflammation in preterm gestation on motor deficits in the newborn, and whether biosynthesis of tetrahydrobiopterin or inflammatory mediators is affected in the fetal brain. METHODS: Pregnant rabbits at 28 days gestation (89% term) were administered either saline or LPS into both endocervical openings. One group underwent spontaneous delivery, and neurobehavioral tests were performed at postnatal day (P) 1 and P11, with some kits being sacrificed at P1 for histological analysis. Another group underwent Cesarean section 24 h after LPS administration. Gene sequences for rabbit biosynthetic enzymes of tetra-hydrobiopterin pathways were determined and analyzed in addition to cytokines, using quantitative real-time polymerase chain reaction. RESULTS: Exposure to 200 μg/kg/mL LPS caused a locomotion deficit and mild hypertonia at P1. By P11, most animals turned into normal-appearing kits. There was no difference in neuronal cell death in the caudate between hypertonic and nonhypertonic kits at P1 (n = 3-5 in each group). Fetal brain GTP cyclohydrolase I was increased, whereas sepiapterin reductase and 6-pyruvoyltetrahydropterin synthase were decreased, 24 h after LPS administration. Neuronal NOS was also increased. Regardless of the position in the uterus or the brain region, expression of TNF-α and TGF-β was decreased, whereas that of IL-1β, IL-6, and IL-8 was increased (n = 3-4 in each group). CONCLUSIONS: This is the first study using an ascending LPS-induced intrauterine inflammation model in rabbits, showing mostly transient hypertonia and mainly locomotor deficits in the kits. Not all proinflammatory cytokines are increased in the fetal brain following LPS administration. Changes in key tetrahydro-biopterin biosynthetic enzymes possibly indicate different effects of the inflammatory insult.

PMID: 31163416