Cerebral Palsy ALLIANCE

Cerebral palsy research news

Monday 8 February 2021

Cerebral Palsy Alliance is delighted to bring you this free weekly bulletin of the latest published research into cerebral palsy. Our organisation is committed to supporting cerebral palsy research worldwide - through information, education, collaboration and funding. Find out more at cerebralpalsy.org.au/our-research

Professor Nadia Badawi AM
Macquarie Group Foundation Chair of Cerebral Palsy

Subscribe to CP Research News

Interventions and Management

1. Relation of Musculoskeletal Strength and Function to Postural Stability in Ambulatory Adults With Cerebral Palsy
Areum K Jensen, Cory E Low, Pooja Pal, Tiffany N Raczynski

Arch Rehabil Res Clin Transl. 2020 Jul 15;2(4):100074. doi: 10.1016/j.arrct.2020.100074. eCollection 2020 Dec.

Objective: To understand the relation of musculoskeletal strength and function to postural stability in ambulatory adults with cerebral palsy (CP) who have already developed muscle atrophy and osteoporosis. Design: Two independent group comparison of adults with CP and those without it. Setting: Laboratory study. Participants: Thirteen adults with CP with sex (9 women: 4 men), age (21-62y), and Gross Motor Function Classification System I-III, and 13 sex-, age-, and body-weight-matched control participants completed our study (N=26). Intervention: Not applicable. Main outcome measure: Bone mineral density (BMD), structural or geometrical deformities (at the proximal region of the femur at the hip joint), and maximal muscular strength (forearm and thigh) were measured. The primary outcome measure was postural stability (balance measured using an automated balance system and a Berg Balance Test). Results: Femoral BMD was significantly lower in the CP group compared to the control group, whereas BMD at lumbar and forearm regions was similar between groups. Geometrical angles, lengths, and diameters at the proximal femur were significantly lower in the CP group. There was a direct relation between BMD in the femoral neck and knee extension peak torque in the control group with no relation in the CP group. Although the control group did not show a relation between muscular strength and balance test, the CP group showed a significant linear relation among improving postural stability with greater levels of muscular strength. Conclusion: There were structural differences at the proximal femur and muscular weakness in adults with CP. In adults with CP, balance appears to be more influenced by structural alterations at the femur than muscular strength compared to the control group.

PMID: 33543099

2. Short and Long-Term Effects of Whole-Body Vibration on Spaticity and Motor Performance in Children With Hemiparetic Cerebral Palsy
Fatih Tekin, Erdoğan Kavlak

Percept Mot Skills. 2021 Feb 3;31512521991095. doi: 10.1177/0031512521991095. Online ahead of print.

This study aimed to investigate the short and long-term effects of Whole-Body Vibration (WBV) therapy on spasticity and motor performance in children with hemiparetic cerebral palsy. We recruited 26 patient participants from among children undergoing conventional physiotherapy in a private rehabilitation center. We randomly assigned 22 participants to equally sized treatment (n = 11) and control (n = 11) groups. We evaluated the participants at the beginning of the study with the Gross Motor Function Measure-88, LEGSys™ Spatio-Temporal Gait Analyzer, SportKAT550™ Portable Computerized Kinesthetic Balance Device and the Modified Ashworth Scale. While children in the treatment group were treated with Compex-Winplate™ to administer WBV in three 15-minute sessions per week for eight weeks, children in the control group

received continued conventional physiotherapy during this period. We then re-evaluated all participants both immediately after the treatment and again 12 weeks after the treatment. Following WBV, both gross motor functions and gait and balance skills were significantly improved ($p < 0.05$), and spasticity in lower and upper extremity muscles was significantly inhibited ($p < 0.05$). These improvements were preserved even after 12 weeks. We conclude that WBV is an effective incremental approach to conventional physiotherapy in children with hemiparetic cerebral palsy for inhibiting spasticity and improving motor performance.

PMID: 33535899

3. Hip displacement in children with cerebral palsy
Sarah J Wordie, Kate E Bugler, Paul R Bessell, James E Robb, Mark S Gaston

Bone Joint J. 2021 Feb;103-B(2):411-414. doi: 10.1302/0301-620X.103B2.BJJ-2020-1528.R1.

Aims: The migration percentage (MP) is one criterion used for surgery in dislocated or displaced hips in children with cerebral palsy (CP). The MP at which a displaced hip can no longer return to normal is unclear. The aim of this paper was to identify the point of no return of the MP through a large population-based study. Methods: All children registered on the Cerebral Palsy Integrated Pathway Scotland surveillance programme undergo regular pelvic radiographs. Any child who had a MP measuring over 35% since the programme's inception in 2013, in at least one hip and at one timepoint, was identified. The national radiography database was then interrogated to identify all pelvic radiographs for each of these children from birth through to the date of analysis. A minimum of a further two available radiographs following the initial measurement of MP ≥ 35% was required for inclusion. Results: A total of 239 children (346 hips) were identified as suitable for analysis at a mean of 6.5 years (2.0 to 14.8) follow-up. In all, 1,485 radiographs taken both prior to and after a hip had a MP ≥ 35% were examined and the MP measured to identify any progression of displacement. Interrogation of the data identified that hips with a MP up to 46% returned to a MP below 40% without intervention, and all hips with a MP equal to or greater than 46% displaced further and the MP did not return to the normal range. Statistical analysis showed the result to be 98% specific with this degree of certainty that hips reaching a MP ≥ 46% would not spontaneously regress. Conclusion: These findings are clinically relevant in showing that it may be reasonable to continue to monitor hips with a MP not exceeding 46%. This threshold will also guide referral for further management of a displacing hip. Cite this article: Bone Joint J 2021;103-B(2):411-414.

PMID: 33517734

4. Factors Influencing Outcomes of the Dysplastic Hip in Nonambulatory Children With Cerebral Palsy
Kristen L Carroll, Alan K Stotts, Glen O Baird, Alyssa L Thorman, Matthew Talmage, Whitney D Moss, Mark L McMulkin, Bruce A MacWilliams

J Pediatr Orthop. 2021 Jan 29. doi: 10.1097/BPO.0000000000001760. Online ahead of print.

Background: Hip dysplasia in the nonambulatory child with spastic cerebral palsy (CP) is a common condition not always effectively treated with conservative measures even when recognized early. Optimal surgical intervention strategies and timing are not clear from previous studies. Contralateral hips with less severe subluxation in these patients also often undergo surgery and little is known of outcomes of these less severe hips. This study aims to clarify treatment factors related to long term success following hip surgery for subluxation in nonambulatory children with CP. Methods: A total of 183 nonambulatory subjects with CP and a minimum of 2-year follow-up were included. All subjects underwent varus rotational osteotomy of the femur; other surgical factors considered were addition of pelvic osteotomy (PO), capsulorrhaphy, and soft tissue releases. Additional factors studied were age at index surgery, sex, and unilateral versus bilateral surgery. Severely subluxated (SS) hips, defined as having >50% migration, were studied separately from contralateral nonsevere hips. Surgeries were deemed successful if final follow-up indicated a migration of <25%; patients with any revision surgeries or >25% migration were categorized as failures. Results: A 60% success rate was found in SS hips and a 68% success rate in nonsevere hips. Age at index surgery did not influence success rates in SS hips. In the nonsevere hips, success was associated with index surgery at older age. The addition of a PO was the only concomitant procedure demonstrated to improve outcomes. In SS hips, those with a successful outcome were 2.5 times more likely to have had a PO. The addition of capsulorrhaphy had a negative effect on the entire group, reducing odds of success to 0.8. No other factors were significant. Conclusions: The findings from this multicenter retrospective study suggest that PO be added to varus rotational osteotomies in patients with severe hip subluxation. Surgery should be undertaken for severe dysplasia without concern for age. The addition of capsulorrhaphy does not improve rate of success. Level of evidence: Level III-retrospective comparative study.

PMID: 33534371

5. Residual lumbar hyperlordosis is associated with worsened hip status 5 years after scoliosis correction in non-ambulant patients with cerebral palsy
Aaron J Buckland, Dainn Woo, H Kerr Graham, Dennis Vasquez-Montes, Patrick Cahill, Thomas J Errico, Paul D Sponseller, Harms Study Group

Spine Deform. 2021 Feb 1. doi: 10.1007/s43390-020-00281-4. Online ahead of print.

Background: Cerebral palsy (CP) is a static encephalopathy with progressive musculoskeletal pathology. Non-ambulant children (GMFCS IV and V) with CP have high rates of spastic hip disease and neuromuscular scoliosis. The effect of spinal fusion and spinal deformity on hip dislocation following total hip arthroplasty has been well studied, however in CP this remains largely unknown. This study aimed to identify factors associated with worsening postoperative hip status (WHS) following corrective spinal fusion in children with GMFCS IV and V CP. Methods: Retrospective review of GMFSC IV and V CP patients in a prospective multicenter database undergoing spinal fusion, with 5 years follow-up. WHS was determined by permutations of baseline (BL), 1 year, 2 years, and 5 years hip status and defined by a change from an enlocated hip at BL that became subluxated, dislocated or resected post-op, or a subluxated hip that became dislocated or resected. Hip status was analyzed against patient demographics, hip position, surgical variables, and coronal and sagittal spinal alignment parameters. Cutoff values for parameters at which the relationship with hip status was significant was determined using receiver operating characteristic curves. Logistic regression determined odds ratios for predictors of WHS. Results: Eighty four patients were included. 37 (44%) had WHS postoperatively. ROC analysis and logistic regression demonstrated that the only spinopelvic alignment parameter that significantly correlated with WHS was lumbar hyperlordosis (T12-L5) > 60° (p = 0.028), OR = 2.77 (CI 1.10-6.94). All patients showed an increase in pre-to-postop LL. Change in LL pre-to-postop was no different between groups (p = 0.318), however the WHS group was more lordotic at BL and postop (pre44°/post58° vs pre32°/post51° in the no change group). Age, sex, Risser, hip position, levels fused, coronal parameters, global sagittal alignment (SVA), thoracic kyphosis, and reoperation were not associated with WHS. Conclusion: Postoperative hyperlordosis(> 60°) is a risk factor for WHS at 5 years after spinal fusion in non-ambulant CP patients. WHS likely relates to anterior pelvic tilt and functional acetabular retroversion due to hyperlordosis, as well as loss of protective lumbopelvic motion causing anterior femoracetabular impingement. Level of evidence: III.

PMID: 33523455

6. Dynamic Surface Exercise Training in Improving Trunk Control and Gross Motor Functions among Children with Quadriplegic Cerebral Palsy: A Single Center, Randomized Controlled Trial
Sravan Reddy, Gandhi Karunanithi Balaji

J Pediatr Neurosci. Jul-Sep 2020;15(3):214-219. doi: 10.4103/jpn.JPN_88_19. Epub 2020 Nov 6.

Background: Dynamic surface provides proprioceptive and vestibular feedback with optimal level of arousal. The activities on unstable environment have greater sensorimotor experiences. There is a lack of evidence examining the benefits of dynamic surface exercise training (DSET) among the children with spastic quadriplegic cerebral palsy (CP). Aim: The aim of the study was to analyze the effect of dynamic surface exercises on trunk control and gross motor functions in children with quadriplegic CP. Materials and methods: A total of 30 children with spastic quadriplegic CP with Gross Motor Function Classification System of levels III and IV were recruited by the simple random sampling method (random number generator) to participate in this randomized controlled study. Recruited children were randomly divided into two groups, DSET group and standard physiotherapy training group. Both the groups received active training program lasting for 60 min, 4 days/week for 6 weeks. Gross Motor Function Measure (GMFM)-88 and Pediatric Balance Scale (PBS) scores were recorded at baseline, and at the end of 6-week post-intervention. Results: Total 30 children with quadriplegic CP with mean age 6.64 ± 2.15 years in experimental group and 6.50 ±1.59 years in control group participated in the study. Experimental group showed a significant difference for GMFM and PBS scores between pre- and post-intervention with $P < 0.005$. A significant difference was observed in GMFM scores between experimental and control group with $P < 0.005$. Conclusion: Six-week dynamic surface exercise therapy along with standard physiotherapy was effective in improving trunk control and gross motor function performance among children with spastic quadriplegic CP aged 6-12 years.

PMID: 33531934

7. The Effect of Vibration Therapy on Walking Endurance in Children and Young People With Cerebral Palsy: Do Age and Gross Motor Function Classification System Matter?
Deborah Telford, Renuka M Vesey, Paul L Hofman, Silmara Gusso

Arch Rehabil Res Clin Transl. 2020 Jun 20;2(3):100068. doi: 10.1016/j.arrct.2020.100068. eCollection 2020 Sep.

Objective: To investigate the effect of age and Gross Motor Function Classification System (GMFCS) level on walking endurance after 20 weeks of vibration therapy in children and young people with cerebral palsy (CP). Design: The study was a clinical trial without control group comparing baseline and postintervention outcomes within participants. Setting: Vibration therapy was performed at school or at home. Assessments took place in a clinical research unit. Participants: Children and young people (N=59) with CP, aged 5-20 years, GMFCS level II, III, or IV, recruited through schools, physiotherapy services, and District Health Board clinics, Auckland, New Zealand. Interventions: Participants performed side-alternating whole-body vibration therapy (WBVT) at 20 Hz and 3-mm amplitude, 9 minutes per day, 4 times per week for 20 weeks. Main outcome measures: Distance walked in the 6-minute walk test (6MWT) was recorded before and after the intervention. Results: Participants baseline results for the 6MWT were lower, independent of age or GMFCS, when compared to non-CP literature. On average, participants walked 12% further in the 6MWT after the intervention (P<.001). There was significant improvement in 6MWT distance in all age groups (5-10y: 16%, P<.001; 11-15y: 10%, P=.001; 16-20y: 13%, P<.001) and all GMFCS levels (level II: 10%, P<.001, level III: 40%, P=.013, level IV: 57%, P=.007). There was a greater percentage improvement in the distance walked in those with GMFCS level III and level IV than level II (P=.049 and P<.001, respectively). Conclusions: WBVT had a beneficial effect on walking endurance in children and young people with CP, independent of age and GMFCS.

PMID: 33543094

8. Immediate effects of short period lower limb ergometer exercise in adolescent and young adult patients with cerebral palsy and spastic diplegia
Junpei Fujimoto, Yasunori Umemoto, Yumi Koike, Kazuya Isida, Keiko Sakamoto, Fumihiro Tajima

J Phys Ther Sci. 2021 Jan;33(1):52-56. doi: 10.1589/jpts.33.52. Epub 2021 Jan 5.

[Purpose] To determine the effects of lower limb ergometer exercise on the spasticity and joint range of motion of the lower extremity and gait function in patients with cerebral palsy and spastic paralysis. [Participants and Methods] This study included 8 participants with cerebral palsy and spastic paralysis (GMFCS levels I to IV) who received care at the outpatient clinic. After a 5-min rest, the lower limb ergometer exercises were performed for 10 min. We measured the participants' arterial blood pressure, pulse rate, passive range of knee joint extension, muscle tone using the Modified Ashworth scale (MAS) and Modified Tardieu scale (MTS), and 10-m walk test (10MWT). Measurements were collected three times (at baseline before exercise, immediately at the end of exercise, and 5 min after exercise during recovery). [Results] The 10-min lower limb ergometer exercise significantly improved the knee joint extension, MAS and MTS scores, and reduced lower extremity spasticity. Furthermore, it significantly increased the range of knee joint extension and decreased the 10MWT score. [Conclusion] The results showed that the 10-minute lower limb ergometer exercise is beneficial in reducing the spasticity of the lower limb muscles and in increasing the range of motion of knee extension in paraplegic patients with cerebral palsy, suggesting that its implementation in young children could prevent spasticity and enhance motor function.

PMID: 33519075

9. The impact of being overweight on the mobility, temporal-spatial and kinematic aspects of gait in children with cerebral palsy
Anita J Mudge, Sinu Thilak, Elizabeth A Wojciechowski, Joshua Burns, Simon P Paget

Obes Res Clin Pract. 2021 Feb 2;S1871-403X(21)00005-3. doi: 10.1016/j.orcp.2021.01.005. Online ahead of print.

Aim: Obesity causes altered gait patterns in typically developing children, but its effect on gait in children with physical disabilities is largely unknown. This study explores associations between body mass index (BMI), functional mobility and gait in children with cerebral palsy (CP). Method: An observational cross-sectional study was conducted using three-dimensional

gait analysis data from 197 children with CP, Gross Motor Functional Classification System (GMFCS) levels I to III. BMI values were categorised using the Centres for Disease Control and Prevention (2000) BMI percentiles, which are specific to age and gender. Regression analyses, with GMFCS level as a covariate, explored associations between BMI category and temporal-spatial, kinematic and functional mobility variables. Analyses included children categorised as healthy weight and overweight/obese only (n = 174), with underweight children excluded (n = 23). Results: 131 children (mean age 10.5 years, SD 3) were categorised as healthy weight and 43 children (mean age 9.6 years, SD 2.5) as overweight or obese. BMI was not associated with most gait variables. Increased double support time, reduced hip extension and increased ankle dorsiflexion were observed in children that were overweight, but most differences were small and of uncertain clinical significance. A lower proportion of overweight children walked independently over 500 m. Conclusion: We found little evidence that BMI has a substantial influence on gait patterns in children with CP but some to suggest it may affect long-distance mobility. Different research strategies are required to improve understanding of relationships between adiposity, strength and function, for effective targeting of interventions to improve mobility.

PMID: 33546995

10. Robotic lower extremity exoskeleton use in a non-ambulatory child with cerebral palsy: a case study
Christa M Diot, Robyn L Thomas, Liliane Raess, James G Wrightson, Elizabeth G Condliffe

Disabil Rehabil Assist Technol. 2021 Feb 4;1-5. doi: 10.1080/17483107.2021.1878296. Online ahead of print.

Purpose: With few treatment options available for non-ambulatory children with cerebral palsy (CP), a robotic lower extremity gait trainer may provide a non-invasive addition to conventional treatment options. This case study investigates the usage and impact of robotic lower extremity gait trainer use in a participant with CP over the initial 3 months of use. Materials and methods: This prospective case study involves a 7-year old female (GMFCS V) with CP (registered clinical trial: NCT04251390). The participant used a Trexo Home robotic gait trainer (Trexo) in the community with assessments occurring in the home and school. Trexo usage and bowel movements (BMs) were tracked daily. Postural control and lower extremity range of motion (ROM) and spasticity were evaluated prior to Trexo use and weekly to biweekly thereafter. Results: The participant used the device an average of 46 min/week, over 3.3 d/week. BM frequency increased from 0.4/d at baseline, to 1.2 (±0.5)/d during Trexo use. There were no diffuse systematic changes in postural stability, ROM or muscle spasticity, but specifically head control and spasticity in the knee flexors had improvements. Conclusions: Data and anecdotal reports suggest that regular use of the Trexo Home robotic gait trainer has positive outcomes on frequency and quality of BMs, and may improve head control, and knee flexor spasticity. Larger controlled studies are needed to evaluate the impacts of Trexo use in children with CP. Implications for Rehabilitation Non-ambulatory children with CP can use and may experience benefits from using a robot-assisted gait trainer (RAGT). Constipation, aspects of balance and focal spasticity may improve.

PMID: 33539714

11. Feasibility of Augmenting Ankle Exoskeleton Walking Performance with Step Length Biofeedback in Individuals with Cerebral Palsy
Ying Fang, Zachary F Lerner

IEEE Trans Neural Syst Rehabil Eng. 2021 Feb 1;PP. doi: 10.1109/TNSRE.2021.3055796. Online ahead of print.

Most people with cerebral palsy (CP) suffer from impaired walking ability and pathological gait patterns. Seeking to improve the effectiveness of gait training in this patient population, this study developed and assessed the feasibility of a real-time biofeedback mechanism to augment untethered ankle exoskeleton-assisted walking performance in individuals with CP. We selected step length as a clinically-relevant gait performance target and utilized a visual interface with live performance scores. An adaptive ankle exoskeleton control algorithm provided assistance proportional to the real-time ankle moment. We assessed lower-extremity gait mechanics and muscle activity in seven ambulatory individuals with CP as they walked with adaptive ankle assistance alone and with ankle assistance plus step-length biofeedback. We achieved our technical validation goal by demonstrating a strong correlation between estimated step length and real step length ($r = 0.77$, $p < 0.001$). We achieved our clinical feasibility goal by demonstrating that biofeedback-plus-assistance resulted in a 14% increase in step length relative to baseline ($p \leq 0.05$), while no difference in step length was observed for assistance alone. Additionally, we observed near immediate improvements in lower-extremity posture, moments, and positive power relative to baseline for biofeedback-plus-

assistance ($p < 0.05$), with none, or more-limited improvements observed for assistance alone. Our findings suggest that providing real-time biofeedback and using step length as the target can be effective for increasing the rate at which individuals with CP improve their gait mechanics when walking with wearable ankle assistance.

PMID: 33523814

12. Is impaired coordination related to match physical load in footballers with cerebral palsy of different sport classes?
Raúl Reina, Aitor Iturricastillo, Daniel Castillo, Alba Roldan, Carlos Toledo, Javier Yanci

J Sports Sci. 2021 Feb 1;1-10. doi: 10.1080/02640414.2021.1880740. Online ahead of print.

Impaired coordination is a characteristic feature in cerebral palsy (CP) football players. This study aimed to determine the relationships of three coordination tests with match physical load during competition of para-footballers from different sport classes. Records from 259 para-footballers from 25 national teams were obtained in four international competitions held in 2018 and 2019. The three coordination tests were conducted prior to competition (i.e., rapid heel-toe, side-stepping, and split jumps), and physical match load was recorded by GPS devices during the real game: i.e., maximum/mean, total distance, distance covered at different speed zones, number of accelerations/decelerations at different intensities, and player load. FT1 and FT3 players have the lowest and highest performance in all the coordination tests, respectively, but inconclusive between-groups differences were obtained ($p=0.022–0.238$). Split jumps and side-stepping tests are associated with the performance of moderate and high accelerations during competition ($r = -0.20–0.71$; $p<0.01$). Significant correlations ($r = 0.36–0.71$; $p<0.01$) were obtained between all the coordination measures. Coordination tests better discriminate those with more severe impairments and some evidence for the validity of the new CP football sport classes is provided. Further research is necessary to identify the portion of the variance in sports performance that coordination explains.

PMID: 33525957

13. Sleep Assessments for Children With Severe Cerebral Palsy: A Scoping Review
Jennifer A Hutson, LeAnn Snow

Review Arch Rehabil Res Clin Transl. 2020 Oct 30;2(4):100087. doi: 10.1016/j.arrct.2020.100087. eCollection 2020 Dec.

Objectives: To identify the sleep-based instruments in postural care intervention research and examine whether the instruments are suitable as postural care outcome measures specifically for children with severe cerebral palsy. Data sources: Investigators searched the electronic databases from 2 university library systems, including OVID Medline, CINAHL, OT Search, Cochrane Database of Systematic Reviews, and Health and Psychosocial Instruments for articles published between 2000 and October 2019. Study selection: The initial search yielded 1928 abstracts. Two independent investigators identified 8 English-language peer-reviewed articles that published postural care intervention study results. Data extraction: Investigators screened the 8 articles and found that 6 included sleep as a primary or secondary intervention outcome. The principal investigator then fully reviewed these 6 publications, recorded their sleep-related instruments, and applied Coster's published guidelines (2013) to analyze the sleep-based instruments' suitability as outcome measures. Data synthesis: Collectively, the 6 studies used 8 distinct measures, 6 of which (actigraphy, Chailey Sleep Questionnaire, Pediatric Sleep Questionnaire, polysomnography, sleep diary, and Sleep Disturbance Scale for Children) underwent analysis. As stand-alone instruments, none completely met criteria for suitability as outcome measures for those with severe cerebral palsy. Conclusions: Combined use of the Sleep Disturbance Scale for Children and actigraphy may be favorable for assessing the sleep-related domains relevant to children with severe cerebral palsy. However, rehabilitation professionals should test sensitivity and specificity to understand the instruments' ability for capturing changes in sleep from postural care intervention.

PMID: 33543110

14. Assessments and Interventions for Sleep Disorders in Infants With or at High Risk for Cerebral Palsy: A Systematic Review
Kelly Tanner, Garey Noritz, Lauren Ayala, Rachel Byrne, Darcy Fehlings, Alison Gehred, Lisa Letzkus, Iona Novak, Nathan Rosenberg, Jilda Vargus-Adams, Sarah Winter, Nathalie L Maitre

Pediatr Neurol. 2020 Nov 3;S0887-8994(20)30348-9. doi: 10.1016/j.pediatrneurol.2020.10.015. Online ahead of print.

Background: Children with cerebral palsy (CP) are five times more likely than typically developing children to have sleep problems, resulting in adverse outcomes for both children and their families. The purpose of this systematic review was to gather current evidence regarding assessments and interventions for sleep in children under age 2 years with or at high risk for CP and integrate these findings with parent preferences. Methods: Five databases (CINAHL, EMBASE, OVID/Medline, SCOPUS, and PsycINFO) were searched. Included articles were screened using preferred reporting items for systematic reviews and meta-analyses guidelines, and quality of the evidence was reviewed using best evidence tools by two independent reviewers at minimum. An online survey was conducted regarding parent preferences through social media channels. Results: Eleven articles met inclusion criteria. Polysomnography emerged as the only high-quality assessment for the population. Three interventions (medical cannabis, surgical interventions, and auditory, tactile, visual, and vestibular stimulations) were identified; however, each only had one study of effectiveness. The quality of evidence for polysomnography was moderate, while the quality and quantity of the evidence regarding interventions was low. Survey respondents indicated that sleep assessments and interventions are highly valued, with caregiver-provided interventions ranked as the most preferable. Conclusions: Further research is needed to validate affordable and feasible sleep assessments compared to polysomnography as the reference standard. In the absence of diagnosis-specific evidence of safety and efficacy of sleep interventions specific to young children with CP, it is conditionally recommended that clinicians follow guidelines for safe sleep interventions for typically developing children.

PMID: 33541756

15. Musculoskeletal Pain Outcomes Pre- and Post Intrathecal Baclofen Pump Implant in Children With Cerebral Palsy: A Prospective Cohort Study

Chantel C Barney, Alyssa M Merbler, Jean Stansbury, Linda E Krach, Michael Partington, Patrick Graupman, Peter D Kim, Debbie Song, Frank J Symons

Arch Rehabil Res Clin Transl. 2020 Mar 9;2(2):100049. doi: 10.1016/j.arrct.2020.100049. eCollection 2020 Jun.

Objective: To characterize musculoskeletal pain intensity, duration, frequency, and interference with activities of daily living in children with cerebral palsy (CP) before and after intrathecal baclofen pump placement. Design: Prospective cohort study. Setting: Children's tertiary hospital. Participants: Participants were children with CP (N=32; 53% male; mean age, 9.9y; age range, 4-17y). The majority of participants had a CP diagnosis of quadriplegia (76%) and relied on wheeled mobility (91%). Interventions: Assessments were completed pre- and post intrathecal baclofen pump implant. Main outcome measures: Because of considerable patient heterogeneity, both pain measures (Brief Pain Inventory, Dalhousie Pain Interview) were completed by proxy (parent) report at the time of the procedure and approximately 6 months after intrathecal baclofen (ITB) pump placement. Results: Prior to implant, 31% of participants were living with constant pain, which reduced to 6% post ITB implant (P<.001). Based on Wilcoxon signed rank tests, pain duration significantly decreased post ITB pump implant (P<.01). Conclusions: This prospective analysis supports the anecdotal and retrospective evidence that musculoskeletal pain decreases in CP following ITB pump implant. The greatest effect appears to be on the duration of pain experience. Pain did not decrease for all individuals, and it would be worth further investigation to better understand the relation between patient characteristics and pain outcomes.

PMID: 33543076

16. Pain and Communication in Children with Cerebral Palsy: Influence on Parents' Perception of Family Impact and Healthcare Satisfaction

Inmaculada Riquelme, Álvaro Sabater-Gárriz, Pedro Montoya

Children (Basel). 2021 Jan 27;8(2):87. doi: 10.3390/children8020087.

Cerebral palsy (CP) is an impacting chronic condition. Concomitant comorbidities such as pain and speech inability may further affect parents' perception of the pathology impact in the family quality of life and the provided care. The objective of this cross-sectional descriptive correlational study was to compare parental reports on family impact and healthcare satisfaction

in children with CP with and without chronic pain and with and without speech ability. Parents of 59 children with CP (age range = 4-18 years) completed several questions about pain and speech ability and two modules of the Pediatric Quality of Life Measurement Model: The PedsQLTM 2.0 Family Impact Module and the PedsQLTM Healthcare Satisfaction Generic Module. Our findings revealed that children's pain slightly impacted family physical health, social health and worry. In children without pain, speech inability increased the perceived health impact. Parents' healthcare satisfaction was barely affected by pain or speech inability, both increasing parents' satisfaction in the professional technical skills and inclusion of family domains on the care plan. In conclusion, pain and speech inability in children with CP can impact family health but not healthcare satisfaction. Regular assessment and intervention in family health is essential for the design of family-centred programs for children with CP.

PMID: 33513751

17. Pediatric dentists can determine chewing performance level in children
Selen Serel Arslan, Cansu Özşin Özler, Numan Demir, Şeyma Öztürk, Meryem Uzamış Tekçiçek, Aynur Ayşe Karaduman

Clin Oral Investig. 2021 Feb 2. doi: 10.1007/s00784-021-03815-w. Online ahead of print.

Objectives: The current study was aimed to verify whether pediatric dentists could determine chewing performance level in children by using Karaduman Chewing Performance Scale (KCPS). Materials and methods: Typical developing children and children with cerebral palsy (CP) who were referred to pediatric dentistry above the age of 2 years were included in the study. The chewing performance level was scored according to KCPS. One experienced physical therapist and three pediatric dentists independently assessed the chewing videos of the children and scored each child's chewing function. The correlation between the KCPS scores of the physical therapist and the pediatric dentists was used for reliability. The agreement between the scorings of the physical therapist and pediatric dentists was assessed using Fleiss kappa statistics. Results: Fifty-four typical developing children and 43 children with CP were included. A strong positive correlation between the KCPS scoring of the physical therapist and pediatric dentists was found ($r=0.911$-0.939, $p<0.001$). An excellent agreement in the KCPS scoring between the physical therapist and the 1st and 3rd dentists ($p<0.001$, κ 0.754-0.763), and a good agreement in the KCPS scoring between the physical therapist and the 2nd dentist was detected ($p<0.001$, κ 0.687). Conclusions: The study results show that the KCPS is reliable for pediatric dentists in determining the chewing performance level in children. Therefore, it could be suggested that pediatric dentists could use the KCPS in their clinical settings and research studies. Clinical relevance: The study may have clinical implications in the evaluation of children with chewing difficulty in dental practice. Clinical trial number: NCT04407455.

PMID: 33528675

18. Acoustic vowel analysis and speech intelligibility in young adult Hebrew speakers: Developmental dysarthria versus typical development
Micalle Carl, Michal Icht

Int J Lang Commun Disord. 2021 Feb 1. doi: 10.1111/1460-6984.12598. Online ahead of print.

Background: Developmental dysarthria is a motor speech impairment commonly characterized by varying levels of reduced speech intelligibility. The relationship between intelligibility deficits and acoustic vowel space among these individuals has long been noted in the literature, with evidence of vowel centralization (e.g., in English and Mandarin). However, the degree to which this centralization occurs and the intelligibility-acoustic relationship is maintained in different vowel systems has yet to be studied thoroughly. In comparison with American English, the Hebrew vowel system is significantly smaller, with a potentially smaller vowel space area, a factor that may impact upon the comparisons of the acoustic vowel space and its correlation with speech intelligibility. Data on vowel space and speech intelligibility are particularly limited for Hebrew speakers with motor speech disorders. Aims: To determine the nature and degree of vowel space centralization in Hebrew-speaking adolescents and young adults with dysarthria, in comparison with typically developing (TD) peers, and to correlate these findings with speech intelligibility scores. Methods & procedures: Adolescents and young adults with developmental dysarthria (secondary to cerebral palsy (CP) and other motor deficits, n = 17) and their TD peers (n = 17) were recorded producing Hebrew corner vowels within single words. For intelligibility assessments, naïve listeners transcribed those words produced by speakers with CP, and intelligibility scores were calculated. Outcomes & results: Acoustic analysis of vowel formants (F1, F2) revealed a centralization of vowel space among speakers with CP for all acoustic metrics of vowel formants, and mainly for the formant centralization ratio (FCR), in comparison with TD peers. Intelligibility scores were correlated

strongly with the FCR metric for speakers with CP. Conclusions & implications: The main results, vowel space centralization for speakers with CP in comparison with TD peers, echo previous cross-linguistic results. The correlation of acoustic results with speech intelligibility carries clinical implications. Taken together, the results contribute to better characterization of the speech production deficit in Hebrew speakers with motor speech disorders. Furthermore, they may guide clinical decision-making and intervention planning to improve speech intelligibility. What this paper adds What is already known on the subject Speech production and intelligibility deficits among individuals with developmental dysarthria (e.g., secondary to CP) are well documented. These deficits have also been correlated with centralization of the acoustic vowel space, although primarily in English speakers. Little is known about the acoustic characteristics of vowels in Hebrew speakers with motor speech disorders, and whether correlations with speech intelligibility are maintained. What this paper adds to existing knowledge This study is the first to describe the acoustic characteristics of vowel space in Hebrew-speaking adolescents and young adults with developmental dysarthria. The results demonstrate a centralization of the acoustic vowel space in comparison with TD peers for all measures, as found in other languages. Correlation between acoustic measures and speech intelligibility scores were also documented. We discuss these results within the context of cross-linguistic comparisons. What are the potential or actual clinical implications of this work? The results confirm the use of objective acoustic measures in the assessment of individuals with motor speech disorders, providing such data for Hebrew-speaking adolescents and young adults. These measures can be used to determine the nature and severity of the speech deficit across languages, may guide intervention planning, as well as measure the effectiveness of intelligibility-based treatment programmes.

PMID: 33522087

19. Motor learning-based activities may improve functional ability in adults with severe cerebral palsy: A controlled pilot study
Helle Hüche Larsen, Rasmus Feld Frisk, Maria Willerslev-Olsen, Jens Bo Nielsen

NeuroRehabilitation. 2021 Jan 23. doi: 10.3233/NRE-201581. Online ahead of print.

Background: Cerebral palsy (CP) is a neurodevelopmental disturbance characterized by impaired control of movement. Function often decreases and 15% of adults are classified as severely affected (Gross Motor Function Classification Scale III-V). Little is known about interventions that aim to improve functional abilities in this population. Objective: To evaluate a 12-week intervention based on motor learning principles on functional ability in adults with severe CP. Methods: 16 adults (36±10 years, GMFCS III-V) were enrolled and divided into an intervention group (Active group) and a standard care group (Control group). Primary outcome measure was Gross Motor Function Measure (GMFM-88). Secondary measures were neurological status. The Active group were measured at baseline, after the intervention and at one-month follow-up. The Control group were measured at baseline and after one month. Results: Analysis showed statistically significant improvement in GMFM-88 for the Active group from baseline to post assessment compared with the Control group (group difference: 5 points, SE 14.5, p = 0.008, CI: 1.2 to 8.7). Improvements were maintained at follow-up. Results from the neurological screening showed no clear tendencies. Conclusions: The study provides support that activities based on motor learning principles may improve gross motor function in adults with severe CP.

PMID: 33523030

20. Effectiveness and safety of pure acupuncture and moxibustion in the treatment of children with cerebral palsy: A protocol for systematic review and meta analysis
Yani Tang, Zhiliang Cao, Yun Xia, Yinghan Liu, Wei Zhang

Medicine (Baltimore). 2021 Jan 29;100(4):e23907. doi: 10.1097/MD.0000000000023907.

Background: Infantile cerebral palsy refers to brain damage in infants and young children during their development, causing brain dysfunction, mainly manifested as dyskinesia, which may be complicated by mental retardation, epilepsy, and bone and joint developmental disorders. Clinical practice shows that acupuncture can effectively treat children with cerebral palsy, but it needs to be proven. This research will systematically evaluate the clinical effectiveness and safety of acupuncture and moxibustion in the treatment of children with cerebral palsy, and provide evidence-based evidence for it. Method: Search the following databases, including CNKI, WANFANG, China Biomedical Database, VIP, PubMed, Embase, the Cochrane Library, Web of Science. The retrieval time is from the establishment of the databases to October 2020, collecting all clinical randomized controlled studies of acupuncture and moxibustion treatment of children with cerebral palsy. Two investigators independently extract and evaluate the data of the included studies, and use RevMan V.5.3 software to conduct meta-analysis

of the included literature. Result: This study evaluates the effectiveness and safety of acupuncture and moxibustion in the treatment of children with cerebral palsy through indicators such as Gross Motor Function Measure Scale, the Modified Ashworth Scale, and so on. Conclusion: This study will provide reliable evidence-based evidence for the clinical application of acupuncture and moxibustion in the treatment of children with cerebral palsy. Ethics and dissemination: Private information from individuals will not be published. This systematic review also does not involve endangering participant rights. Ethical approval was not required. The results may be published in a peer-reviewed journal or disseminated at relevant conferences. Osf registration number: DOI 10.17605/OSF.IO/7GUF5.

PMID: 33530188

21. Psychiatric disorders in adults with cerebral palsy
Carly A McMorris, Johanna Lake, Kristin Dobranowski, Caitlin McGarry, Elizabeth Lin, Drew Wilton, Yona Lunsky, Robert Balogh

Res Dev Disabil. 2021 Jan 29;111:103859. doi: 10.1016/j.ridd.2021.103859. Online ahead of print.

Background: Cerebral palsy (CP) is one of the most common neurological conditions in childhood. Individuals with CP often experience various secondary conditions, including intellectual disability (ID), medical conditions, and psychiatric issues. A large number of youth with CP have psychiatric disorders; however, few studies have examined the prevalence of psychiatric issues in adults with CP at the population-level. Aims: To investigate the prevalence and co-occurrence of psychiatric disorders at the population-level in adults with CP only, and adults with CP and ID. Method and procedures: Using clinical information from seven Canadian data sources, we conducted a retrospective cross-sectional analysis of adults with CP, with and without ID. Outcomes and results: Adults with CP were more likely than the general population to have a psychiatric diagnosis, independent of ID status. All psychiatric disorders were more common in individuals with CP than the general population, with the exception of addiction related disorders. In most cases, having an ID substantially increased the risk of having a psychiatric disorder. Conclusions: Adults with CP are at heightened risk for experiencing psychiatric disorders. Current findings highlight the important role health care providers play in screening for psychiatric issues in individuals with CP.

PMID: 33524738

22. Cerebral palsy in twins and higher multiple births: a Europe-Australia population-based study
Elodie Sellier, Shona Goldsmith, Sarah McIntyre, Oliver Perra, Gija Rackauskaite, Nadia Badawi, Asma Fares, Hayley Smithers-Sheedy, Surveillance Of Cerebral Palsy Europe Group And The Australian Cerebral Palsy Register Group

Dev Med Child Neurol. 2021 Feb 2. doi: 10.1111/dmcn.14827. Online ahead of print.

Aim: To describe the birth prevalence, temporal trends, and clinical outcomes of twins, triplets, or quadruplets with cerebral palsy (CP). Method: This was a cross-sectional study using data for twins, triplets, and quadruplets with prenatally or perinatally acquired CP and pooled from the Surveillance of Cerebral Palsy in Europe network (born 1992-2009) and Australian Cerebral Palsy Register (born 1993-2009). Children were at least 4 years old at time of registration. Children born in regions with population ascertainment and available denominator data were included in prevalence calculations (n=1033 twins, 81 triplets, and 11 quadruplets). Clinical data from children registered in all participating registers were described, including 2163 twins (56% male), 187 triplets (59% male), and 20 quadruplets (45% male). Results: The birth prevalence of CP was higher with increasing plurality (twins 6.5 per 1000 live births [95% confidence interval {CI} 6.1-6.9], triplets 17.1 [95% CI 13.6-21.2], quadruplets 50.7 [95% CI 25.6-88.9]); however, prevalence by gestational age was similar across all pluralities. Between 1992-1994 and 2007-2009, prevalence of CP among twins declined (p=0.001) but prevalence of CP among triplets did not change significantly over time (p=0.55). The distributions of Gross Motor Function Classification System, epilepsy, and impairments of intellect, vision, and hearing were similar regardless of plurality. Interpretation: The data combined from two CP register networks indicated that triplets and quadruplets had increased risk of CP compared to twins. The higher prevalence of CP in triplets and quadruplets is due to their higher risk of preterm birth. Prevalence of CP among twins significantly declined in Europe and Australia. Clinical outcomes were similar for all multiple births.

PMID: 33533028

23. Cerebral Palsy Research Network Clinical Registry: Methodology and Baseline Report
Paul Gross, Mary Gannotti, Amy Bailes, Susan D Horn, Jacob Kean, Unni G Narayanan, Jerry Oakes, Garey Noritz, Cerebral Palsy Research Network

Arch Rehabil Res Clin Transl. 2020 Apr 19;2(3):100054. doi: 10.1016/j.arrct.2020.100054. eCollection 2020 Sep.

Objective: To apply practice-based evidence to clinical management of cerebral palsy (CP). The process of establishing purpose, structure, logistics, and elements of a multi-institutional registry and the baseline characteristics of initial enrollees are reported. Design: A consensus-building process among consumers, clinicians, and researchers used a participatory action process. Setting: Community, hospitals, and universities. Participants: More than 100 clinicians, researchers, and consumers and more than 1858 enrollees in the registry. Main outcome measures: Not applicable. Results: Consensus was that the purpose of registry was to (1) quantify practice variation, (2) facilitate quality improvement (QI), and (3) perform comparative effectiveness research (CER). Collecting data during routine clinical care using the electronic medical record was determined to be a sustainable plan for data acquisition and management. Clinicians from multiple disciplines defined salient characteristics of individuals and interventions for the registry elements. The registry was central to the clinical research network, and a leadership structure was created. A leading electronic health record platform adopted the registry elements. Twenty-four sites have initiated the data collection process and agreed to export data to the registry. Currently 12 are collecting data. Number of enrollees and characteristics were similar to other population registers. Conclusions: This is the first multi-institutional CP registry that contains the patient and treatment characteristics needed for QI and CER. The Cerebral Palsy Research Network registry elements are implemented in a versatile electronic platform and minimize burden to clinicians. The resultant registry is available for any institution to participate and is growing rapidly.

PMID: 33543081

24. Participation in Social Roles of Adolescents With Cerebral Palsy: Exploring Accomplishment and Satisfaction
Dirk-Wouter Smits, Marloes van Gorp, Leontien van Wely, Johannes Verheijden, Jeanine Voorman, Sophie Wintels, Joyce van der Cruijsen, Marjolijn Ketelaar, PERRIN-PiP study group

Arch Rehabil Res Clin Transl. 2019 Aug 22;1(3-4):100021. doi: 10.1016/j.arrct.2019.100021. eCollection 2019 Dec.

Objective: To explore participation in social roles of adolescents (aged 12-18y) with cerebral palsy (CP), in terms of satisfaction compared with accomplishment. Design: Cohort study as part of a prospective longitudinal research program. Setting: Clinic. Participants: Participants were adolescents (N=45; 58% male, mean age 15y 6mo) with CP at levels I-II (88%) and III-IV-V (12%) of the Gross Motor Function Classification System. Interventions: Not applicable. Main outcome measures: Accomplishment (0-9 scale; with score <8 "having difficulties") and satisfaction (1-5 scale; with score 3 "neutral") were assessed using the Life-Habits questionnaire, on 6 domains (Responsibilities, Interpersonal relationships, Community life, Education, Employment, Recreation). Per domain, we analyzed scatterplots of accomplishment vs satisfaction. Additionally, we compared determinant-models (including factors of CP, activity, person, and environment) using regression analysis. Results: For accomplishment, mean scores were <8.00 except for Interpersonal relationships. For satisfaction, mean scores varied between 3.85 and 4.34. Overall, individuals with similar levels of accomplishment showed large ranges in their levels of satisfaction, which was expressed by low explained variances, especially on Education (6%). Furthermore, different sets of determinants were found for accomplishment (predominantly CP factors) compared with satisfaction (predominantly environment factors). Conclusions: This study revealed a dissociation between participation accomplishment and satisfaction with participation among adolescents with CP. For practice and research, we recommend not only to focus on accomplishment but also, if not mainly, on satisfaction.

PMID: 33543052

25. Examining Conductive Education: Linking Science, Theory, and Intervention
Roberta O'Shea, Mary Jones, Katie Lightfoot

Arch Rehabil Res Clin Transl. 2020 Aug 22;2(4):100077. doi: 10.1016/j.arrct.2020.100077. eCollection 2020 Dec.

Recognized in many European countries and Canada as a valid form of therapeutic and educational rehabilitation, conductive

education (CE) emphasizes cognitive and motor learning principles for movement reeducation. This article illustrates how CE incorporates motor control and motor learning theories in conjunction with unique facilitation concepts, including rhythmic intention, task series, tailored low-tech equipment, and traditional facilitation concepts such as developmental sequence, manual facilitations, and multimodal interventions. Uniquely, CE brings together task series practice and learning, including a lying program, sitting program, standing program, and walking program, along with activities of daily living within a group treatment model. The conductor uses cadence and rhythmic intention to encourage movement exploration in a scripted plan of care. The participants are active learners and use CE slatted equipment to help support movements. Full participation, to the best of the learners' ability, is realized with activity modifications made by the conductor. Increased motor control arises through repetition, practice, functional context, and sensory feedback that provide guidance for intention and voluntary movement. Motor control and motor learning theories are foundational principles of CE. Individuals with neurologic injuries, including cerebral palsy, stroke syndrome, Parkinson disease, and traumatic brain injury, can benefit from CE. To date, although research studies cannot objectively compare one person's movement skills with another's, new research surrounding motor control and motor learning illustrates and supports the principles and practice of CE. CE is an educational therapy model for teaching and developing new movement skills for individuals with neurologic impairments. This article connects the current science of movement and describes the unique principles involved with CE delivery as an intervention for individuals with neurologic impairments.

PMID: 33543102

26. Molecular Diagnostic Yield of Exome Sequencing in Patients With Cerebral Palsy
Andrés Moreno-De-Luca, Francisca Millan, Denis R Pesacreta, Houda Z Elloumi, Matthew T Oetjens, Claire Teigen, Karen E Wain, Julie Scuffins, Scott M Myers, Rebecca I Torene, Vladimir G Gainullin, Kevin Arvai, H Lester Kirchner, David H Ledbetter, Kyle Retterer, Christa L Martin

JAMA. 2021 Feb 2;325(5):467-475. doi: 10.1001/jama.2020.26148.

Importance: Cerebral palsy is a common neurodevelopmental disorder affecting movement and posture that often co-occurs with other neurodevelopmental disorders. Individual cases of cerebral palsy are often attributed to birth asphyxia; however, recent studies indicate that asphyxia accounts for less than 10% of cerebral palsy cases. Objective: To determine the molecular diagnostic yield of exome sequencing (prevalence of pathogenic and likely pathogenic variants) in individuals with cerebral palsy. Design, setting, and participants: A retrospective cohort study of patients with cerebral palsy that included a clinical laboratory referral cohort with data accrued between 2012 and 2018 and a health care-based cohort with data accrued between 2007 and 2017. Exposures: Exome sequencing with copy number variant detection. Main outcomes and measures: The primary outcome was the molecular diagnostic yield of exome sequencing. Results: Among 1345 patients from the clinical laboratory referral cohort, the median age was 8.8 years (interquartile range, 4.4-14.7 years; range, 0.1-66 years) and 601 (45%) were female. Among 181 patients in the health care-based cohort, the median age was 41.9 years (interquartile range, 28.0-59.6 years; range, 4.8-89 years) and 96 (53%) were female. The molecular diagnostic yield of exome sequencing was 32.7% (95% CI, 30.2%-35.2%) in the clinical laboratory referral cohort and 10.5% (95% CI, 6.0%-15.0%) in the health care-based cohort. The molecular diagnostic yield ranged from 11.2% (95% CI, 6.4%-16.2%) for patients without intellectual disability, epilepsy, or autism spectrum disorder to 32.9% (95% CI, 25.7%-40.1%) for patients with all 3 comorbidities. Pathogenic and likely pathogenic variants were identified in 229 genes (29.5% of 1526 patients); 86 genes were mutated in 2 or more patients (20.1% of 1526 patients) and 10 genes with mutations were independently identified in both cohorts (2.9% of 1526 patients). Conclusions and relevance: Among 2 cohorts of patients with cerebral palsy who underwent exome sequencing, the prevalence of pathogenic and likely pathogenic variants was 32.7% in a cohort that predominantly consisted of pediatric patients and 10.5% in a cohort that predominantly consisted of adult patients. Further research is needed to understand the clinical implications of these findings.

PMID: 33528536

27. Gene expressions in cerebral palsy subjects reveal structural and functional changes in the gastrocnemius muscle that are closely associated with passive muscle stiffness
Jessica Pingel, Marie-Louise Kampmann, Jeppe Dyrberg Andersen, Christian Wong, Simon Døssing, Claus Børsting, Jens Bo Nielsen

Cell Tissue Res. 2021 Jan 30. doi: 10.1007/s00441-020-03399-z. Online ahead of print.

Cerebral palsy (CP) is a non-progressive motor disorder that affects posture and gait due to contracture development. The purpose of this study is to analyze a possible relation between muscle stiffness and gene expression levels in muscle tissue of children with CP. Next-generation sequencing (NGS) of gene transcripts was carried out in muscle biopsies from gastrocnemius muscle (n = 13 children with CP and n = 13 typical developed (TD) children). Passive stiffness of the ankle plantarflexors was measured. Structural changes of the basement membranes and the sarcomere length were measured. Twelve pre-defined gene target sub-categories of muscle function, structure and metabolism showed significant differences between muscle tissue of CP and TD children. Passive stiffness was significantly correlated to gene expression levels of HSPG2 ($p = 0.02$; $R2 = 0.67$), PRELP ($p = 0.002$; $R2 = 0.84$), RYR3 ($p = 0.04$; $R2 = 0.66$), C COL5A3 ($p = 0.0007$; $R2 = 0.88$), ASPH ($p = 0.002$; $R2 = 0.82$) and COL4A6 ($p = 0.03$; $R2 = 0.97$). Morphological differences in the basement membrane were observed between children with CP and TD children. The sarcomere length was significantly increased in children with CP when compared with TD ($p = 0.04$). These findings show that gene targets in the categories: calcium handling, basement membrane and collagens, were significantly correlated to passive muscle stiffness. A Reactome pathway analysis showed that pathways involved in DNA repair, ECM proteoglycans and ion homeostasis were amongst the most upregulated pathways in CP, while pathways involved in collagen fibril crosslinking, collagen fibril assembly and collagen turnover were amongst the most downregulated pathways when compared with TD children. These results underline that contracture formation and motor impairment in CP is an interplay between multiple factors.

PMID: 33515289

28. HIF1A polymorphisms do not modify the risk of epilepsy nor cerebral palsy after neonatal hypoxic-ischemic encephalopathy
Eva Kukec, Katja Goričar, Vita Dolžan, Zvonka Rener Primec

Brain Res. 2021 Jan 27;147281. doi: 10.1016/j.brainres.2021.147281. Online ahead of print.

Purpose: Hypoxic-ischemic encephalopathy (HIE) remains the major cause of cerebral palsy and epilepsy in developed countries. Hypoxia-inducible factor 1 alpha (HIF-1α) is the key mediator of oxygen homoeostasis. The aim of this study was to investigate whether hypoxia-inducible factor 1 subunit alpha (HIF1A) functional polymorphisms are associated with the risk of epilepsy, drug-resistant epilepsy, and cerebral palsy after neonatal HIE. Methods: The study included 139 healthy controls and 229 patients with epilepsy and/or cerebral palsy, of which 95 had perinatal HIE. Genomic DNA isolated from buccal swabs or peripheral blood were genotyped for HIF1A rs11549465 and rs11549467 using PCR based methods. Results: The investigated HIF1A polymorphisms did not influence the risk of epilepsy and its drug-resistance nor cerebral palsy after neonatal HIE (all $p > 0.05$). Clinical characteristics of patients were significantly associated with neurological deficits after HIE. Conclusion: This study found no statistically significant association of HIF1A rs11549465 and rs11549467 with the development of epilepsy and its drug-resistance, as well as cerebral palsy, after neonatal HIE.

PMID: 33515534

29. Ophthalmic phenotypes associated with biallelic loss-of-function PCDH12 variants
Francesca Mattioli, Norine Voisin, Eglė Preikšaitienė, Irina Kozlovskaja, Vaidutis Kučinskas, Alexandre Reymond

Case Reports Am J Med Genet A. 2021 Feb 2. doi: 10.1002/ajmg.a.62098. Online ahead of print.

Individuals carrying biallelic loss-of-function mutations in PCDH12 have been reported with three different conditions: the diencephalic-mesencephalic junction dysplasia syndrome 1 (DMJDS1), a disorder characterized by global developmental delay, microcephaly, dystonia, and a midbrain malformation at the diencephalic-mesencephalic junction; cerebral palsy combined with a neurodevelopmental disorder; and cerebellar ataxia with retinopathy. We report an additional patient carrying a homozygous PCDH12 frameshift, whose anamnesis combines the most recurrent DMJDS1 clinical features, that is, global developmental delay, microcephaly, and ataxia, with exudative vitreoretinopathy. This case and previously published DMJDS1 patients presenting with nonspecific visual impairments and ophthalmic disorders suggest that ophthalmic alterations are an integral part of clinical features associated with PCDH12 loss-of-function.

PMID: 33527719

30. Altered cytokine responses in children with cerebral palsy: pathogenesis and novel therapies
Mark R Schleiss

Dev Med Child Neurol. 2021 Feb 2. doi: 10.1111/dmcn.14821. Online ahead of print.

PMID: 33533040

31. Neurological Manifestations of Congenital Cytomegalovirus Infection at a Tertiary Care Centre from Southern India
Vykuntaraju K Gowda, Preeti Kulhalli, Dhananjaya K Vamyanmane

J Neurosci Rural Pract. 2021 Jan;12(1):133-136. doi: 10.1055/s-0040-1721557. Epub 2021 Jan 29.

Background Cytomegalovirus (CMV) is a ubiquitous herpes virus. It is the most common congenital viral infection. Data on congenital CMV in India are lacking and hence the present study was undertaken. Objectives The aim of the study is to evaluate the clinical and radiological profile of neurological manifestations of congenital CMV infections in tertiary care hospital. Methods This is a retrospective chart review of the clinical and laboratory profile of congenital CMV infections presenting from January 2018 to February 2020 to a tertiary care hospital in Southern India. Details of clinical profile, serological and neuroimaging data were obtained and analyzed. Results A total of 42 cases with female preponderance (57%) were reported during the study period. The mean age of presentation was 2.9 years. Clinical features were developmental delay (81%), microcephaly (93%), seizures (33%), intrauterine growth restriction (19%), neonatal encephalopathy (10%), anemia (9%), jaundice (10%), hepato-splenomegaly (7%), and eye abnormalities (14%). Antenatal maternal fever was reported by 12%. Sensorineural hearing loss was present in 57%. Neuroimaging showed periventricular calcification (79%), cerebral atrophy (69%), ventricular dilatation (55%), malformations (26%), dysmyelination (12%), and temporal lobe cysts (5%). CMV -immunoglobulin-M positivity was seen in 14 cases (33%), urinary polymerase chain reaction for CMV was positive in 21 cases (50%), and clinical diagnosis was done in seven cases (16%). Conclusion Common findings in congenital CMV are microcephaly, developmental delay, seizures, anemia, and sensorineural hearing loss. Common neuroimaging findings are periventricular calcification, cerebral atrophy, malformation, white matter signal changes, and cysts. CMV can mimic like cerebral palsy, malformations of the brain, demyelinating disorders, and calcified leukoencephalopathies like Aicardi-Goutières syndrome.

PMID: 33531772

32. Neurodevelopmental profiles of children with unilateral cerebral palsy associated with middle cerebral artery and periventricular venous infarctions
Darcy Fehlings, Pradeep Krishnan, Renee-Marie Ragguett, Gabrielle deVeber, Jan Willem Gorter, Carolyn Hunt, Marie Kim, Ronit Mesterman, Anna McCormick, CP-NET Group

Dev Med Child Neurol. 2021 Jan 31. doi: 10.1111/dmcn.14818. Online ahead of print.

Aim: To compare the neurodevelopment of children with unilateral cerebral palsy (CP) with middle cerebral artery (MCA) and periventricular venous infarctions (PVIs). Method: In this cross-sectional study, children with unilateral CP completed a neurological exam, unimanual Quality of Upper Extremity Skills Test, hand usage questionnaires, and IQ test. Neuroimaging was obtained from health records. Results: Two hundred and forty-five participants with unilateral CP had neuroimaging (151 [61.9%] male, ages 2-18y, median=7y 6mo, interquartile range [IQR]=6y 7mo, with 93.6% in Gross Motor Function Classification System level I/II and 78.8% in Manual Ability Classification System level I/II). Ninety-seven (39.6%) had MCA injuries and 106 (43.3%) had periventricular white matter injuries, of which 48 (45.3%) were PVIs. Median Quality of Upper Extremity Skills Test for the MCA group was 49.2 (IQR=55.8), PVI 79.9 (IQR=23.6) (Mann-Whitney U=988.50, $p<0.001$). Bimanual hand usage (Children's Hand-use Experience Questionnaire) (Mann-Whitney U=425, $p<0.001$) and light touch (odds ratio=9.12, 95% confidence interval 1.28-400.76, Fisher's exact test $p=0.017$) were lower in the MCA compared to the PVI group. Full-scale IQ median centile score for the MCA group was 18.0 (IQR=35.5) and 50.0 (IQR=30.0) for the PVI group (Mann-Whitney U=382, $p<0.001$). Interpretation: Children with unilateral CP and MCA injuries demonstrated lower hand function and usage, decreased light touch, and lower IQs compared to the PVI group. This study aids in defining rehabilitation needs informed by brain injury patterns.

PMID: 33521966

33. Evaluating transcranial magnetic stimulation (TMS) induced electric fields in pediatric stroke
Kathleen E Mantell, Ellen N Sutter, Sina Shirinpour, Samuel T Nemanich, Daniel H Lench, Bernadette T Gillick, Alexander Opitz

Neuroimage Clin. 2021 Jan 13;29:102563. doi: 10.1016/j.nicl.2021.102563. Online ahead of print.

Transcranial magnetic stimulation (TMS) is an increasingly popular tool for stroke rehabilitation. Consequently, researchers have started to explore the use of TMS in pediatric stroke. However, the application of TMS in a developing brain with pathologies comes with a unique set of challenges. The effect of TMS-induced electric fields has not been explored in children with stroke lesions. Here, we used finite element method (FEM) modeling to study how the electric field strength is affected by the presence of a lesion. We created individual realistic head models from MRIs (n = 6) of children with unilateral cerebral palsy due to perinatal stroke. We conducted TMS electric field simulations for coil locations over lesioned and non-lesioned hemispheres. We found that the presence of a lesion can strongly affect the electric field distribution. On the group level, the mean electric field strength did not differ between lesioned and non-lesioned hemispheres but exhibited a greater variability in the lesioned hemisphere. Other factors such as coil-to-cortex distance have a strong influence on the TMS electric field even in the presence of lesions. Our study has important implications for the delivery of TMS in children with brain lesions with respect to TMS dosing and coil placement.

PMID: 33516935

34. Changes of Structural Brain Network Following Repetitive Transcranial Magnetic Stimulation in Children With Bilateral Spastic Cerebral Palsy: A Diffusion Tensor Imaging Study
Wenxin Zhang, Shang Zhang, Min Zhu, Jian Tang, Xiaoke Zhao, Ying Wang, Yuting Liu, Ling Zhang, Hong Xu

Front Pediatr. 2021 Jan 15;8:617548. doi: 10.3389/fped.2020.617548. eCollection 2020.

Introduction: Bilateral spastic cerebral palsy (BSCP) is the most common subtype of cerebral palsy (CP), which is characterized by various motor and cognitive impairments, as well as emotional instability. However, the neural basis of these problems and how repetitive transcranial magnetic stimulation (rTMS) can make potential impacts on the disrupted structural brain network in BSCP remain unclear. This study was aimed to explore the topological characteristics of the structural brain network in BSCP following the treatment of rTMS. Methods: Fourteen children with BSCP underwent 4 weeks of TMS and 15 matched healthy children (HC) were enrolled. Diffusion tensor imaging (DTI) data were acquired from children with bilateral spastic cerebral palsy before treatment (CP1), children with bilateral spastic cerebral palsy following treatment (CP2) and HC. The graph theory analysis was applied to construct the structural brain network. Then nodal clustering coefficient (C i) and shortest path length (L i) were measured and compared among groups. Results: Brain regions with significant group differences in C i were located in the left precental gyrus, middle frontal gyrus, calcarine fissure, cuneus, lingual gyrus, postcentral gyrus, inferior parietal gyri, angular gyrus, precuneus, paracentral lobule and the right inferior frontal gyrus (triangular part), insula, posterior cingulate gyrus, precuneus, paracentral lobule, pallidum. In addition, significant differences were detected in the L i of the left precental gyrus, lingual gyrus, superior occipital gyrus, middle occipital gyrus, superior parietal gyrus, precuneus and the right median cingulate gyrus, posterior cingulate gyrus, hippocampus, putamen, thalamus. Post hoc t-test revealed that the CP2 group exhibited increased C i in the right inferior frontal gyrus, pallidum and decreased L i in the right putamen, thalamus when compared with the CP1 group. Conclusion: Significant differences of node-level metrics were found in various brain regions of BSCP, which indicated a disruption in structural brain connectivity in BSCP. The alterations of the structural brain network provided a basis for understanding of the pathophysiological mechanisms of motor and cognitive impairments in BSCP. Moreover, the right inferior frontal gyrus, putamen, thalamus could potentially be biomarkers for predicting the efficacy of TMS.

PMID: 33520901

35. Early developmental screening and intervention for high-risk neonates - From research to clinical benefits
Alicia Jane Spittle, Peter John Anderson, Sarah Jane Tapawan, Lex William Doyle, Jeanie Ling Yoong Cheong

Review Semin Fetal Neonatal Med. 2021 Jan 29;101203. doi: 10.1016/j.siny.2021.101203. Online ahead of print.

With advances in neonatal care there has been an increase in survival rates for infants born very preterm and/or with complex needs, such as those who require major surgery, who may not have survived decades ago. Despite advances in survival, these infants remain at high-risk for a range of neurodevelopmental delays and/or impairments including motor, cognitive and emotional/behavioural challenges. Research has improved our ability to identify which infants are at high-risk of developmental delay and/or impairments, and there is mounting evidence that early interventions can improve outcomes of these infants. However, clinical practice varies throughout the world regarding recommendations for developmental screening. Moreover, intervention, when available, is often not commenced early enough in development. Given limited resources, those infants most at risk of developmental impairments and their families should be targeted, with further research needed on the cost-effectiveness of surveillance and early interventions.

PMID: 33547000

36. A Retrospective Cohort Study on Mortality and Neurodevelopmental Outcomes of Preterm Very Low Birth Weight Infants Born to Mothers with Hypertensive Disorders of Pregnancy
Yumi Kono, Naohiro Yonemoto, Hidehiko Nakanishi, Shigeharu Hosono, Shinya Hirano, Satoshi Kusuda, Masanori Fujimura, Neonatal Research Network Japan

Am J Perinatol. 2021 Feb 3. doi: 10.1055/s-0041-1722874. Online ahead of print.

Objective: We examined the effects of maternal hypertensive disorders of pregnancy (HDP) on the mortality and neurodevelopmental outcomes in preterm very low birth weight (VLBW) infants (BW ≤1,500 g) based on their intrauterine growth status and gestational age (GA). Study design: We included singleton VLBW infants born at <32 weeks' gestation registered in the Neonatal Research Network Japan database. The composite outcomes including death, cerebral palsy (CP), and developmental delay (DD) at 3 years of age were retrospectively compared among three groups: appropriate for GA (AGA) infants of mothers with and without HDP (H-AGA and N-AGA) and small for GA (SGA) infants of mothers with HDP (H-SGA). The adjusted odds ratios (AOR) and 95% confidence intervals (CI) stratified by the groups of every two gestational weeks were calculated after adjusting for the center, year of birth, sex, maternal age, maternal diabetes, antenatal steroid use, clinical chorioamnionitis, premature rupture of membranes, non-life-threatening congenital anomalies, and GA. Results: Of 19,323 eligible infants, outcomes were evaluated in 10,192 infants: 683 were H-AGA, 1,719 were H-SGA, and 7,790 were N-AGA. Between H-AGA and N-AGA, no significant difference was observed in the risk for death, CP, or DD in any GA groups. H-AGA had a lower risk for death, CP, or DD than H-SGA in the 24 to 25 weeks group (AOR: 0.434, 95% CI: 0.202-0.930). The odds for death, CP, or DD of H-SGA against N-AGA were found to be higher in the 24 to 25 weeks (AOR: 2.558, 95% CI: 1.558-3.272) and 26 to 27 weeks (AOR: 1.898, 95% CI: 1.427-2.526) groups, but lower in the 30 to 31 weeks group (AOR: 0.518, 95% CI: 0.335-0.800). Conclusion: There was a lack of follow-up data; however, the outcomes of liveborn preterm VLBW infants of mothers with HDP depended on their intrauterine growth status and GA at birth.

PMID: 33535243

37. Growth and survival characteristics of spa mice
Joline E Brandenburg, Matthew J Fogarty, Gary C Sieck

Animal Model Exp Med. 2020 Oct 10;3(4):319-324. doi: 10.1002/ame2.12137. eCollection 2020 Dec.

Characterization of growth and survival of mice displaying early onset hypertonic symptoms is critical as these animals are important for research investigating mechanisms and treatments of pediatric conditions associated with hypertonia, such as cerebral palsy. Currently, most animal models of cerebral palsy reproduce risk factors for developing this condition, with most failing to develop the physical symptoms or failing to survive in the postnatal period. The B6.Cg-Glrbspa /J (Gly receptor mutation) transgenic mouse (spa mouse), displays symptoms of early onset hypertonia, though little has been reported on growth and survival, with no reports of growth and survival since genotyping became available. We found that the majority of spa mice display symptoms by P14-P16. Of mice surviving to weaning, only ~9% were spa mice. By weaning age, spa mice had significantly lower weights than their heterozygote and wild-type littermates. Of mice that died after weaning and prior to use in experiments or being culled, 48% were spa mice. The poor growth and decreased survival of spa mice across multiple developmental and adult ages resembled the varied survival rates observed in humans with mild or severe cerebral palsy. The understanding of the expected survival of these mice is helpful for planning breeding and animal numbers for experiments. Due to the symptoms and timing of symptom onset, spa mice will be valuable in uncovering mechanisms and long-term effects of

early onset hypertonia in order to move toward interventions for these conditions.

PMID: 33532707

38. Prematurity Rates During the Coronavirus Disease 2019 (COVID-19) Pandemic Lockdown in Melbourne, Australia
Alexia Matheson, Claire J McGannon, Atul Malhotra, Kirsten R Palmer, Alice E Stewart, Euan M Wallace, Ben W Mol, Ryan J Hodges, Daniel L Rolnik

Obstet Gynecol. 2021 Feb 4. doi: 10.1097/AOG.0000000000004236. Online ahead of print.

PMID: 33543904

39. Creatine Metabolism in Female Reproduction, Pregnancy and Newborn Health
Anna Maria Muccini, Nhi T Tran, Deborah L de Guingand, Mamatha Philip, Paul A Della Gatta, Robert Galinsky, Larry S Sherman, Meredith A Kelleher, Kirsten R Palmer, Mary J Berry, David W Walker, Rod J Snow, Stacey J Ellery

Review Nutrients. 2021 Feb 2;13(2):490. doi: 10.3390/nu13020490.

Creatine metabolism is an important component of cellular energy homeostasis. Via the creatine kinase circuit, creatine derived from our diet or synthesized endogenously provides spatial and temporal maintenance of intracellular adenosine triphosphate (ATP) production; this is particularly important for cells with high or fluctuating energy demands. The use of this circuit by tissues within the female reproductive system, as well as the placenta and the developing fetus during pregnancy is apparent throughout the literature, with some studies linking perturbations in creatine metabolism to reduced fertility and poor pregnancy outcomes. Maternal dietary creatine supplementation during pregnancy as a safeguard against hypoxia-induced perinatal injury, particularly that of the brain, has also been widely studied in pre-clinical in vitro and small animal models. However, there is still no consensus on whether creatine is essential for successful reproduction. This review consolidates the available literature on creatine metabolism in female reproduction, pregnancy and the early neonatal period. Creatine metabolism is discussed in relation to cellular bioenergetics and de novo synthesis, as well as the potential to use dietary creatine in a reproductive setting. We highlight the apparent knowledge gaps and the research "road forward" to understand, and then utilize, creatine to improve reproductive health and perinatal outcomes.

PMID: 33540766

40. Supporting caregivers of children born prematurely in the development of language: A scoping review
Roxanne Belanger, Dominique Leroux, Pascal Lefebvre

Paediatr Child Health. 2019 Oct 21;26(1):e17-e24. doi: 10.1093/pch/pxz124. eCollection 2021 Feb.

Background: Infants born prematurely can display impairments that negatively impact the early years of their development. Compared to their peers born at term, preterm children have higher risks of cerebral palsy, sensory deficits, learning disabilities, cognitive and language deficits, as well as difficulties related to attention and behaviour. Following discharge, parents of preterm children are often supported through neonatal follow-up programs or by community health care practitioners. Through assessment and consultation, professionals foster parental resilience by teaching them about their child's development. Research shows a large volume of literature on improving outcomes for preterm infants, but less attention has been given to the impact and potential importance of education of parents regarding the care they provide from the home. Objective: A scoping review was completed to determine the best practices for early intervention in premature children regarding the development of language skills during the preschool years. Methods: The review followed the guidelines for the Preferred Reporting Items for Systematic reviews and Meta-Analyses (PRISMA). Results: Four general themes emerged from the review and included the importance of providing (1) parental training in the care of an infant born prematurely during neonatal intensive care unit stay; (2) education on the development of language and the importance of parental responsiveness; (3) provision of activities to support child language learning; and (4) overall and ongoing monitoring and support by qualified health professionals. Conclusions: The conclusions drawn will provide guidance to health care professionals regarding the education of parents on best practices for stimulating language development in their child.

PMID: 33542775

41. [The application of neonatal intensive care unit main caregiver ability evaluation index system in premature infants with hypoxic ischemic encephalopathy][Article in Chinese]
Xiaoyan Zhao, Jing Gao, Xinxin Kang, Shaohua Yan, Ya Li, Rui Huang

Zhonghua Wei Zhong Bing Ji Jiu Yi Xue. 2020 Dec;32(12):1487-1490. doi: 10.3760/cma.j.cn121430-20200714-00521.

Objective: To explore the effect of main caregiver ability evaluation index system (MCAEIS) in neonatal intensive care unit (NICU) in the treatment of hypoxic-ischemic encephalopathy (HIE). Methods: One hundred and eight cases of HIE preterm infants treated in NICU of the First Hospital of Hebei Medical University from August 2018 to August 2019 were retrospectively analyzed. The infants were divided into routine nursing control group and MCAEIS group, 54 cases each. The control group recieved NICU routine care, and the MCAEIS group was given care by NICU MCAEIS. The nursing ability of the main caregivers in the two groups was evaluated by self-made caregiver ability self-assessment questionnaire; the neurological function was assessed by neonatal behavioral neurological score (NBNA) at discharge and one month after discharge; the incidence of neurological sequelae mental retardation [development quotient (DQ) < 75], epilepsy, cerebral palsy and so on were observed. Results: There were no significant differences in the scores of nursing knowledge, nursing technology or nursing ability between the two groups at admission, but the above scores were significantly improved in the two groups at discharge, and the scores of nursing knowledge, nursing technology and nursing ability of main caregivers in the MCAEIS group were significantly higher than those in the routine nursing control group (nursing knowledge score: 29.84±3.47 vs. 20.83±3.94,nursing technology: 31.47±4.56 vs. 25.12±4.18,nursing ability: 17.28±2.39 vs.12.83±4.78), with significant differences (all $P < 0.05$). One month after discharge, the neurological function score in MCAEIS group was significantly higher than that of routine nursing control group (39.67±3.76 vs. 35.87±5.71, $P < 0.001$). The incidence of neurological sequelae in MCAEIS group was significantly lower than that in the routine nursing control group [1.85% (1/54) vs. 12.96% (7/54), $P = 0.031$]. Conclusions: NICU MCAEIS could scientifically and systematically evaluate the nursing ability of the main caregivers, improve the nursing ability of children, and provide a reference for clinical nursing, which is worthy of widely application.

PMID: 33541502

42. Cost burden and net monetary benefit loss of neonatal hypoglycaemia
Matthew J Glasgow, Richard Edlin, Jane E Harding

BMC Health Serv Res. 2021 Feb 5;21(1):121. doi: 10.1186/s12913-021-06098-9.

Background: Neonatal hypoglycaemia is a common but treatable metabolic disorder that affects newborn infants and which, if not identified and treated adequately, may result in neurological sequelae that persist for the lifetime of the patient. The long-term financial and quality-of-life burden of neonatal hypoglycaemia has not been previously examined. Methods: We assessed the postnatal hospital and long-term costs associated with neonatal hypoglycaemia over 80 year and 18 year time horizons, using a health-system perspective and assessing impact on quality of life using quality-adjusted life year (QALYs). A decision analytic model was used to represent key outcomes in the presence and absence of neonatal hypoglycaemia. Results: The chance of developing one of the outcomes of neonatal hypoglycaemia in our model (cerebral palsy, learning disabilities, seizures, vision disorders) was 24.03% in subjects who experienced neonatal hypoglycaemia and 3.56% in those who do did not. Over an 80 year time horizon a subject who experienced neonatal hypoglycaemia had a combined hospital and post-discharge cost of NZ$72,000 due to the outcomes modelled, which is NZ$66,000 greater than a subject without neonatal hypoglycaemia. The net monetary benefit lost due to neonatal hypoglycaemia, using a value per QALY of NZ$43,000, is NZ$180,000 over an 80 year time horizon. Conclusions: Even under the most conservative of estimates, neonatal hypoglycaemia contributes a significant financial burden to the health system both during childhood and over a lifetime. The combination of direct costs and loss of quality of life due to neonatal hypoglycaemia means that this condition warrants further research to focus on prevention and effective treatment.

PMID: 33546675

43. Measuring Early Problem-Solving in Young Children with Motor Delays: A Validation Study

Rebecca M Molinini, Natalie A Koziol, Tanya Tripathi, Regina T Harbourne, Sarah Westcott McCoy, Michele A Lobo, James Bovaird, Stacey C Dusing

Phys Occup Ther Pediatr. 2021 Feb 1;1-19. doi: 10.1080/01942638.2020.1865501. Online ahead of print.

Aim: There is a lack of evidence-based tools for measuring problem-solving in young children with motor delays. The purpose of this study was to evaluate the construct validity and responsiveness of the Assessment of Problem-Solving in Play. Methods: 125 young children (10.72, SD 2.62 months) with mild, moderate, and severe motor delays were assessed with the Bayley Scales of Infant and Toddler Development, Third Edition Cognitive Scale and the Assessment of Problem-Solving in Play up to 4 times over 12 months. The baseline and change over time assessment scores were compared. Results: The Assessment of Problem-Solving in Play was strongly, positively correlated with the Bayley Scales of Infant and Toddler Development, Third Edition Cognitive Scale raw scores at baseline (r=.83, p<.001) and for changes in scores across time (r=.64, p<.001). On average, participants demonstrated positive change in problem-solving scores across time. Participants with severe motor delay scored lower at baseline and changed less as compared to other participants. Conclusions: Results provide evidence for the construct validity and responsiveness of the Assessment of Problem-Solving in Play scores in quantifying problem-solving in young children with motor delays 7-27 months of age.

PMID: 33517815

Prevention and Cure

44. Translating antenatal magnesium sulphate neuroprotection for infants born <28 weeks' gestation into practice: A geographical cohort study

Lex W Doyle, Alicia J Spittle, Joy E Olsen, Amanda Kwong, Rosemarie A Boland, Katherine J Lee, Peter J Anderson, Jeanie L Y Cheong, Victorian Infant Collaborative Study Group

Aust N Z J Obstet Gynaecol. 2021 Feb 2. doi: 10.1111/ajo.13301. Online ahead of print.

Background: Magnesium sulphate was introduced for fetal neuroprotection in Australia in 2010. The aim of this study was to determine how often antenatal magnesium sulphate is used currently and its association with cerebral palsy in children born <28 weeks' gestation. Materials and methods: Participants comprised all survivors born <28 weeks' gestational age in the state of Victoria in 2016-17, and earlier, in 1991-92, 1997, 2005. Rates of cerebral palsy, diagnosed at two years for the 2016-17 cohort, and at eight years in the earlier cohorts, were compared across eras. Within 2016-17, the proportions of children exposed to antenatal magnesium sulphate were determined, and rates of cerebral palsy were compared between those with and without exposure to magnesium sulphate. Results: Overall, cerebral palsy was present in 6% (11/171) of survivors born in 2016-17, compared with 12% (62/499) of survivors born in the three earlier eras (odds ratio (OR) 0.48, 95% confidence interval (CI) 0.25-0.94; P = 0.032). Data were available for 213/215 (99%) survivors born in 2016-17, of whom 147 (69%) received magnesium sulphate. Data on cerebral palsy at two years were available for 171 (80%) survivors with magnesium data. Cerebral palsy was present in 5/125 (4%) children exposed to magnesium sulphate and in 6/46 (13%) of those not exposed (OR 0.28, 95% CI 0.08-0.96; P = 0.043). Conclusions: Antenatal magnesium sulphate is being translated into clinical practice for infants born <28 weeks' gestation, but there is room for improvement. It is associated with lower rates of cerebral palsy in survivors.

PMID: 33528040