Cerebral palsy research news

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Interventions and Management

1. Major complications following surgical correction of spine deformity in 257 patients with cerebral palsy
Burt Yaszay, Carrie E Bartley, Paul D Sponseller, Mark Abel, Patrick J Cahill, Suken A Shah, Firoz Miyanji, Amer F Samdani, Carlie Daquino, Peter O Newton

Spine Deform. 2020 Jul 27;1-8. doi: 10.1007/s43390-020-00165-7. Online ahead of print.

Study design: Observational. Objectives: To report on the rate of major complications following spinal fusion and instrumentation to treat spinal deformity in patients with cerebral palsy (CP). Understanding the risk of major complications following the surgical treatment of spine deformities in patients with CP is critical. Methods: A prospectively collected, multicenter database of patients with CP who had surgical correction of their spinal deformity (scoliosis or kyphosis) was reviewed for all major complications. Patients with ≥ 2 year follow-up or who died ≤ 2 years of surgery were included. A complication was defined as major if it resulted in reoperation, re-admission to the hospital, prolongation of the hospital stay, was considered life-threatening, or resulted in residual disability. Overall complication and revision rates were calculated for the perioperative (Peri-op; occurring ≤ 90 days postoperative) and delayed postoperative (Delayed; > 90 days) time periods. Results: Two hundred and fifty-seven patients met inclusion. Seventy-eight (30%) patients had a major complication, 18 (7%) had > 1. There were 92 (36%) major complications; 64 (24.9%) occurred Peri-op. The most common Peri-op complications were wound (n = 16, 6.2%) and pulmonary issues (n = 28, 10.9%), specifically deep infections (n = 12, 4.7%) and prolonged ventilator support (n = 21, 8.2%). Delayed complications (n = 28, 10.9%) were primarily deep infections (n = 8, 3.1%) and instrumentation-related (n = 6, 2.3%). There were 42 additional surgeries for an overall unplanned return to the operating room rate of 16% (Peri-op: 8.6%, Delayed: 7.8%). Thirty-six (14.0%) reoperations were spine related surgeries (wound or instrumentation-related). Eleven (4.3%) patients died between 3 months to 5.6 years postoperatively; 4 occurred ≤ 1 year of surgery. Two deaths were directly related to the spinal deformity surgery. Conclusion: Spinal deformity surgery in CP patients with greater than 2 years of follow-up have a postoperative major complication rate of 36% with a spine-related reoperation rate of 14.0%. Level of evidence: Therapeutic-IV.

PMID: 32720268

2. Cervical myelopathy in cerebral palsy
Mengxian Jia, Jinwei Ying, Honglin Teng

Dev Med Child Neurol. 2020 Jul 25. doi: 10.1111/dmcn.14641. Online ahead of print.

PMID: 32710638

3. Effects of whole-body vibration combined with action observation on gross motor function, balance, and gait in children with spastic cerebral palsy: a preliminary study
Youngmin Jung, Eun-Jung Chung, Hye-Lim Chun, Byoung-Hee Lee

J Exerc Rehabil. 2020 Jun 30;16(3):249-257. doi: 10.12965/jer.2040136.068. eCollection 2020 Jun.

This study evaluated the effects of whole-body vibration (WBV) com-bined with action observation on gross motor function, balance, and gait in children with spastic cerebral palsy. The participants were randomized into the WBV combined with action observation (WBVAO) group (n=7) and the WBV group (n=7). The WBVAO group received WBV combined with action observation training, and the WBV group received WBV training for 4 weeks. Both groups received 30 min of training a day, 3 times a week. All participants completed the 5 times sit to stand (FTSTS) test, Timed Up and Go (TUG) test, pediatric reach test, 10-m walk test, 6-min walk test (6MWT), and timed up and down stair (TUDS) test before and after the training intervention. Moreover, the participant's Gross Motor Function Measure (GMFM) and Pediatric Balance Scale (PBS) scores were assessed. Both the WBVAO and WBV groups demonstrated significant increases in the scores of FTSTS test, GMFM (D and E dimensions), PBS, TUG test, 6MWT, and TUDS test. The WBVAO group improved more significantly compared to the WBV group based on the scores of the FTSTS test, GMFM (D dimension), PBS, 6MWT, and TUDS test. WBV combined with action observation improved lower extremity functional strength, gross motor function, and balance and gait in children with cerebral palsy. These results suggest that WBV combined with action observation is both feasible and suitable for individuals with cerebral palsy.

PMID: 32724782

4. Body Representations in Children with Cerebral Palsy
Antonella Di Vita, Maria Cristina Cinelli, Simona Raimo, Maddalena Boccia, Stefano Buratin, Paola Gentili, Maria Teresa Inzitari, Teresa Iona, Marco Iosa, Daniela Morelli, Francesco Ruggeri, Giuseppina Russo, Cecilia Guariglia, Liana Palermo

Brain Sci. 2020 Jul 28;10(8):E490. doi: 10.3390/brainsci10080490.

We constantly process top-down and bottom-up inputs concerning our own body that interact to form body representations (BR). Even if some evidence showed BR deficits in children with cerebral palsy, a systematic study that evaluates different kinds of BR in these children, taking into account the possible presence of a general deficit affecting non-body mental representations, is currently lacking. Here we aimed at investigating BR (i.e., Body Semantics, Body Structural Representation and Body Schema) in children with cerebral palsy (CP) taking into account performance in tasks involving body stimuli and performance in tasks involving non-body stimuli. Thirty-three CP (age range: 5-12 years) were compared with a group of 103 typically-developing children (TDC), matched for age and sex. 63.64% of children with CP showed a very poor performance in body representation processing. Present data also show alterations in different body representations in CP in specific developmental stages. In particular, CP and TDC performances did not differ between 5 to 7 years old, whereas CP between 8 and 12 years old showed deficits in the Body Structural Representation and Body Schema but not in Body Semantics. These findings revealed the importance of taking into account the overall development of cognitive domains when investigating specific stimuli processing in children who do not present a typical development and were discussed in terms of their clinical implications.

PMID: 32731526

5. Structural Brain Lesions and Gait Pathology in Children With Spastic Cerebral Palsy
Eirini Papageorgiou, Nathalie De Beukelaer, Cristina Simon-Martinez, Lisa Mailleux, Anja Van Campenhout, Kaat Desloovere, Els Ortibus

Front Hum Neurosci. 2020 Jul 9;14:275. doi: 10.3389/fnhum.2020.00275. eCollection 2020.

The interaction between brain damage and motor function is not yet fully understood in children with spastic cerebral palsy (CP). Therefore, a semi-quantitative MRI (sqMRI) scale was used to explore whether identified brain lesions related to functional abilities and gait pathology in this population. A retrospective cohort of ambulatory children with spastic CP was selected [N = 104; 52 bilateral (bCP) and 52 unilateral (uCP)]. Extent and location-specific scores were defined according to

the sqMRI scale guidelines. The gross motor function classification system (GMFCS), the gait profile score (GPS), GPSs per motion plane, gait variable scores (GVS) and multiple-joint (MJ) gait patterns were related to brain lesion scores. In all groups, the global total brain scores correlated to the GPS (total: r s = 0.404, p ≤ 0.001; bCP: r s = 0.335, p ≤ 0.05; uCP: r s = 0.493, p ≤ 0.001). The global total hemispheric scores correlated to the GMFCS (total: r s = 0.392, p ≤ 0.001; bCP: r s = 0.316, p ≤ 0.05; uCP: r s = 0.331, p ≤ 0.05). The laterality scores of the hemispheres in the total group correlated negatively to the GMFCS level (r s = -0.523, p ≤ 0.001) and the GVS-knee sagittal (r s = -0.311, p ≤ 0.01). Lesion location, for the total group demonstrated positive correlations between parietal lobe involvement and the GPS (r s = 0.321, p ≤ 0.001) and between periventricular layer damage and the GMFCS (r s = 0.348, p ≤ 0.001). Involvement of the anterior part of the corpus callosum (CC) was associated with the GVS-hip sagittal in all groups (total: r pb = 0.495, p ≤ 0.001; bCP: r pb = 0.357, p ≤ 0.05; uCP: r pb = 0.641, p ≤ 0.001). The global total hemispheric and laterality of the hemispheres scores differentiated between the minor and both the extension (p ≤ 0.001 and p ≤ 0.001) and flexion (p = 0.016 and p = 0.013, respectively) MJ patterns in the total group. Maximal periventricular involvement and CC intactness were associated with extension patterns (p ≤ 0.05 and p ≤ 0.001, respectively). Current findings demonstrated relationships between brain structure and motor function as well as pathological gait, in this cohort of children with CP. These results might facilitate the timely identification of gait pathology and, ultimately, guide individualized treatment planning of gait impairments in children with CP.

PMID: 32733223

6. Muscle Synergies During Walking in Children With Cerebral Palsy: A Systematic Review
Annike Bekius, Margit M Bach, Marjolein M van der Krogt, Ralph de Vries, Annemieke I Buizer, Nadia Dominici

Front Physiol. 2020 Jul 2;11:632. doi: 10.3389/fphys.2020.00632. eCollection 2020.

Background: Walking problems in children with cerebral palsy (CP) can in part be explained by limited selective motor control. Muscle synergy analysis is increasingly used to quantify altered neuromuscular control during walking. The early brain injury in children with CP may lead to a different development of muscle synergies compared to typically developing (TD) children, which might characterize the abnormal walking patterns. Objective: The overarching aim of this review is to give an overview of the existing studies investigating muscle synergies during walking in children with CP compared to TD children. The main focus is on how muscle synergies differ between children with CP and TD children, and we examine the potential of muscle synergies as a measure to quantify and predict treatment outcomes. Methods: Bibliographic databases were searched by two independent reviewers up to 22 April 2019. Studies were included if the focus was on muscle synergies of the lower limbs during walking, obtained by a matrix factorization algorithm, in children with CP. Results: The majority (n = 12) of the 16 included studies found that children with CP recruited fewer muscle synergies during walking compared to TD children, and several studies (n = 8) showed that either the spatial or temporal structure of the muscle synergies differed between children with CP and TD children. Variability within and between subjects was larger in children with CP than in TD children, especially in more involved children. Muscle synergy characteristics before treatments to improve walking function could predict treatment outcomes (n = 3). Only minimal changes in synergies were found after treatment. Conclusions: The findings in this systematic review support the idea that children with CP use a simpler motor control strategy compared to TD children. The use of muscle synergy analysis as a clinical tool to quantify altered neuromuscular control and predict clinical outcomes seems promising. Further investigation on this topic is necessary, and the use of muscle synergies as a target for development of novel therapies in children with CP could be explored.

PMID: 32714199

7. Anatomical basis of a safe mini-invasive technique for lengthening of the anterior gastrocnemius aponeurosis
Simone Moroni, Alejandro Fernández-Gibello, Gabriel Camunas Nieves, Ruben Montes, Marit Zwierzina, Teresa Vazquez, Maria Garcia-Escudero, Fabrice Duparc, Bernhard Moriggl, Marko Konschake

Surg Radiol Anat. 2020 Jul 23. doi: 10.1007/s00276-020-02536-1. Online ahead of print.

Background: The surgical procedure itself of lengthening the gastrocnemius muscle aponeurosis is performed to treat multiple musculoskeletal, neurological and metabolical pathologies related to a gastro-soleus unit contracture such as plantar fasciitis, Achilles tendinopathy, metatarsalgia, cerebral palsy, or diabetic foot ulcerations. Therefore, the aim of our research was to prove the effectiveness and safety of a new ultrasound-guided surgery-technique for the lengthening of the anterior gastrocnemius muscle aponeurosis, the "GIAR"- technique: the gastrocnemius-intramuscular aponeurosis release. Methods and results: An ultrasound-guided surgical GIAR on ten fresh-frozen specimens (10 donors, 8 male, 2 females, 5 left and 5 right)

was performed. Exclusion criteria of the donated bodies to science were BMI above 35 (impaired ultrasound echogenicity), signs of traumas in the ankle and crural region, a history of ankle or foot ischemic vascular disorder, surgery or space-occupying mass lesions. The surgical procedures were performed by two podiatric surgeons with more than 6 years of experience in ultrasound-guided procedures. The anterior gastrocnemius muscle aponeurosis was entirely transected in 10 over 10 specimens, with a mean portal length of 2 mm (± 1 mm). The mean gain at the ankle joint ROM after the GIAR was 7.9° (± 1.1°). No damages of important anatomical structures could be found. Conclusion: Results of this study indicate that our novel ultrasound-guided surgery for the lengthening of the anterior gastrocnemius muscle aponeurosis (GIAR) might be an effective and safe procedure.

PMID: 32705404

8. The utility of shear wave elastography in the evaluation of muscle stiffness in patients with cerebral palsy after botulinum toxin A injection
Nurullah Dağ, Mahi Nur Cerit, Halit Nahit Şendur, Murat Zinnuroğlu, Bilge Nur Muşmal, Emetullah Cindil, Suna Özhan Oktar

J Med Ultrason (2001). 2020 Jul 24. doi: 10.1007/s10396-020-01042-6. Online ahead of print.

Purpose: The first aim of this study was to evaluate changes in the stiffness of the medial gastrocnemius muscle (GM) after a botulinum toxin A (BoNT-A) injection in children with cerebral palsy (CP) using shear wave elastography (SWE). We also wanted to investigate the usability of SWE for evaluating spasticity in a clinical setting. The second aim of this study was to show how treatment of the gastrocnemius muscle spasticity caused a change in the elasticity of the anterior tibial (TA) muscle. Methods: Twenty-four pediatric patients diagnosed with a spastic type of CP, who were scheduled to receive a BoNT-A injection in the gastrocnemius muscle, were included in the study. There was a total of 43 lower extremities to evaluate, and muscle stiffness was measured before the injection and a month post injection using SWE. The physiatrist evaluated muscle spasticity using the Modified Ashworth Scale (MAS) and the Modified Tardieu Scale at about the same time. Results: SWE values of the GM (pre-BoNT-A: 45.9 ± 6.5 kPa, post-BoNT-A: 25.0 ± 5.7 kPa) decreased significantly post BoNT-A injection ($P < 0.01$). SWE measurements of the GM had positive correlations with MAS, V1X, V3X, and R2-R1 ($P < 0.01$); and negative correlations with R2 and R1 ($P < 0.05$). SWE values of the TA muscle (pre: 36.9 ± 7.9 kPa, post: 28.4 ± 5.2 kPa) decreased significantly ($P < 0.01$). Conclusion: Quantitative measurement of muscle stiffness using SWE may provide important information for the evaluation of spasticity and treatment efficiency in pediatric CP patients.

PMID: 32705628

9. Room for improvement: metrological properties of passive muscle-tendon stiffness measures in children with cerebral palsy
Clément Boulard, Raphaël Gross, Vincent Gautheron, Thomas Lapole

Eur J Appl Physiol. 2020 Jul 26. doi: 10.1007/s00421-020-04434-1. Online ahead of print.

PMID: 32715392

10. Sensomotoric Orthoses, Ankle-Foot Orthoses, and Children with Cerebral Palsy: The Bigger Picture
Clare MacFarlane, Robin Orr, Wayne Hing

Children (Basel). 2020 Jul 24;7(8):E82. doi: 10.3390/children7080082.

Ankle-foot orthoses (AFOs) and sensomotoric orthoses (SMotOs) are two-clinically relevant, yet under researched-types of lower limb orthoses used in children with cerebral palsy (CP). Quality of life is a state of complete physical, mental and social well-being and not merely the absence of disease or infirmity. Evaluating the effect of these two types of orthoses on quality of life in children with CP has not been reported on. The aim of this case study series was to synthesise and enrich the volume of evidence reported to inform real world applications of SMotO use in children with CP. Participants recruited were children with CP who performed the Berg Balance Scale, Timed Up-and-Go, the Gross Motor Function Measure and/or the Edinburgh

Visual Gait Score in AFOs, SMotOs and barefoot where able. Qualitative data included videos of gait, a questionnaire and pedographs. Eight participants completed 39 quantitative and six qualitative measures, with the Edinburgh Visual Gait Score (EVGS) reporting the highest response. A general improvement was seen in gross motor skills and gait when wearing the SMotOs compared to AFOs and some parents reported that SMotOs were preferred. The reader is able to correlate the quantitative results with the qualitative evidence presented.

PMID: 32722251

11. [Translation and cross-cultural adaptation of the Gross Motor Function Measure to the Spanish population of children with cerebral palsy][Article in Spanish]
M Ferre-Fernández, M A Murcia-González, J Ríos-Díaz

Rev Neurol. 2020 Sep 1;71(5):177-185. doi: 10.33588/rn.7105.2020087.

Introduction: The Gross Motor Function Measure (GMFM) is a measure designed to assess changes in gross motor function over time in children with cerebral palsy. It is an observation instrument, valid, reliable and responsive, widely used both in research and in clinical practice. Aim: To perform the translation and cross-cultural adaptation to the Spanish population of the GMFM. Subjects and methods: The forward-backward translation methodology was used, subjecting the resulting versions to a qualitative analysis of equivalence. Both the score sheet and the instructions were translated through strategies of omission, incorporation, substitution of words or contribution of examples. In addition, understandability, applicability and feasibility were assessed through a pilot study in which assessors and subjects with a heterogeneous profile participated. Results: The items that generated the most difficulty were those that included clinical terms or expressions whose use is not considered natural or equivalent in the Spanish language. Although 57% and 58% of the items of forward and backward translation, respectively, were classified as «partially equivalent», no correction was necessary since the modifications made came from the cultural and linguistic adaptation of the items to the Spanish population. Conclusions: The Spanish version maintains the highest degree of equivalence concerning the original English version and is understandable by all professionals regardless of their professional experience or geographic origin.

PMID: 32729109

12. Machine Learning to Quantify Physical Activity in Children with Cerebral Palsy: Comparison of Group, Group-Personalized, and Fully-Personalized Activity Classification Models
Matthew N Ahmadi, Margaret E O'Neil, Emmah Baque, Roslyn N Boyd, Stewart G Trost

Sensors (Basel). 2020 Jul 17;20(14):E3976. doi: 10.3390/s20143976.

Pattern recognition methodologies, such as those utilizing machine learning (ML) approaches, have the potential to improve the accuracy and versatility of accelerometer-based assessments of physical activity (PA). Children with cerebral palsy (CP) exhibit significant heterogeneity in relation to impairment and activity limitations; however, studies conducted to date have implemented "one-size fits all" group (G) models. Group-personalized (GP) models specific to the Gross Motor Function Classification (GMFCS) level and fully-personalized (FP) models trained on individual data may provide more accurate assessments of PA; however, these approaches have not been investigated in children with CP. In this study, 38 children classified at GMFCS I to III completed laboratory trials and a simulated free-living protocol while wearing an ActiGraph GT3X+ on the wrist, hip, and ankle. Activities were classified as sedentary, standing utilitarian movements, or walking. In the cross-validation, FP random forest classifiers (99.0-99.3%) exhibited a significantly higher accuracy than G (80.9-94.7%) and GP classifiers (78.7-94.1%), with the largest differential observed in children at GMFCS III. When evaluated under free-living conditions, all model types exhibited significant declines in accuracy, with FP models outperforming G and GP models in GMFCS levels I and II, but not III. Future studies should evaluate the comparative accuracy of personalized models trained on free-living accelerometer data.

PMID: 32708963

13. Effect of lidocaine iontophoresis combined with exercise intervention on gait and spasticity in children with spastic

hemiplegic cerebral palsy: A randomized controlled trial
Fatma A Hegazy, Emad A Aboelnasr, Yasser T Salem

NeuroRehabilitation. 2020 Jul 21. doi: 10.3233/NRE-203152. Online ahead of print.

Background: Spasticity is a common impairment seen in children with cerebral palsy (CP) and may interfere with functional performance and effective walking pattern. Lidocaine iontophoresis is effective for reducing muscle spasticity in adults. Purpose: To investigate the effect of lidocaine epinephrine iontophoresis combined with exercises on gait and spasticity in children with spastic hemiplegic cerebral palsy (HCP). Methods: Thirty children with spastic HCP aged 4-6 (5.20±0.32) years were randomly assigned to the experimental group (n = 15) and control group (n = 15). Children in both groups received one hour of exercises, three times a week for three months. Children in the experimental group received 2% lidocaine iontophoresis immediately before the exercises. The lidocaine iontophoresis was delivered for 20 minutes (1mA/min). Spatio-temporal gait parameters were assessed within one week before and after the intervention using 3D motion analysis. Surface electromyography was used to assess muscle tone using H/M ratio of the soleus muscle. Results: There was no difference between groups at baseline. Post-intervention, the experimental group showed significant improvements when compared to the control group for gait speed (p = 0.03), stride length (p = 0.04), cadence (p = 0.0001), cycle time (p = 0.0001), and H/M ratio (p = 0.02). Conclusion: Lidocaine iontophoresis combined with exercises was effective in improving gait spatiotemporal parameters and reducing spasticity in children with CP.

PMID: 32716326

14. The Formula for Health and Well-Being in Individuals With Cerebral Palsy: Cross-Sectional Data on Physical Activity, Sleep, and Nutrition
Patrick G McPhee, Olaf Verschuren, Mark D Peterson, Ada Tang, Jan Willem Gorter

Ann Rehabil Med. 2020 Jul 28. doi: 10.5535/arm.19156. Online ahead of print.

Objective: To determine physical activity, sleep, and nutrition patterns in individuals with cerebral palsy (CP) and investigate the association of Gross Motor Function Classification System (GMFCS) and age with these health behaviors. Methods: A cross-sectional study was conducted in an outpatient setting. Participants included adolescents and adults with CP (n=28; GMFCS level I-V; mean age 35.1±14.4 years). An Exercise Questionnaire or Leisure Time Physical Activity Questionnaire was used to measure physical activity in adolescents and adults, respectively. Sleep quality was measured using the Pittsburgh Sleep Quality Index (PSQI). An adapted version of the PrimeScreen questionnaire was used to assess nutrition. Linear regression analyses were performed to investigate the association between GMFCS and age with physical activity, sleep, and nutrition. Results: The average total physical activity was 29.2±30.0 min/day. Seventy-five percent of the participants had poor sleep quality (PSQI score >5). Seventy-one percent reported "fair" eating behaviors; none reported "excellent" eating behaviors. Neither GMFCS nor age were significantly associated with PSQI score, PrimeScreen score, or total physical activity. A negative correlation existed between sleep quantity (hr/night) and PSQI score (r=-0.66, p=0.01). Conclusion: The triad of health components, consisting of physical activity, sleep, and nutrition, was not associated with GMFCS or age in our sample of 28 individuals with CP, suggesting that these three health behaviors should be assessed during clinical encounters of CP in adolescents and adults at all levels of the GMFCS.

PMID: 32721989

15. Communication disorders in young children with cerebral palsy
Lindsay Pennington, Mona Dave, Jennifer Rudd, Mary Jo Cooley Hidecker, Katy Caynes, Mark S Pearce

Dev Med Child Neurol. 2020 Jul 30. doi: 10.1111/dmcn.14635. Online ahead of print.

Aim: To test the prediction of communication disorder severity at 5 years of age from characteristics at 2 years for children with cerebral palsy (CP) whose communication is giving cause for concern. Method: In this cohort study, 77 children (52 males; 25 females) with communication difficulties and CP were visited at home at 2 (mean 2y 4mo; SD 3mo) and 5 (mean 5y 5mo; SD 4mo) years of age. Information on the type and distribution of motor disorder, seizures, gross and fine motor function, hearing, and vision were collected from medical notes. Non-verbal cognition, language comprehension, language expression,

spoken vocabulary, and methods of communication were assessed directly at age 2 years. At 5 years, communication and speech function were rated using the Communication Function Classification System (CFCS), Functional Communication Classification System (FCCS), and Viking Speech Scale (VSS). Results: In multivariable regression models, CP type, Gross Motor Function Classification System level, vision, the amount of speech understood by strangers, non-verbal cognition, and number of consonants produced at age 2 years predicted the CFCS level at age 5 years (R2 =0.54). CP type, Manual Ability Classification System level, amount of speech understood, vision, and number of consonants predicted the FCCS level (R2 =0.49). CP type, amount of speech understood by strangers, and number of consonants predicted the VSS level (R2 =0.50). Interpretation: Characteristics at 2 years of age predict communication and speech performance at 5 years, and should inform referral to speech and language therapy.

PMID: 32729634

16. Assessing language comprehension in motor impaired children needing AAC: validity and reliability of the Norwegian version of the receptive language test C-BiLLT
Sara Ida Fiske, Anne Lise Haddeland, Ingvild Skipar, Jael N Bootsma, Johanna J Geytenbeek, Kristine Stadskleiv

Augment Altern Commun. 2020 Jul 24;1-12. doi: 10.1080/07434618.2020.1786857. Online ahead of print.

Children with severe motor impairments who need augmentative and alternative communication (AAC) comprise a heterogeneous group with wide variability in cognitive functioning. Assessment of language comprehension will help find the best possible communication solution for each child, but there is a lack of appropriate instruments. This study investigates the reliability and validity of the Norwegian version of the spoken language comprehension test C-BiLLT (computer-based instrument for low motor language testing) - the C-BiLLT-Nor - and whether response modality influences test results. The participants were 238 children with typical development aged 1;2 to 7;10 (years/months) who were assessed with the C-BiLLT -Nor and tests of language comprehension and non-verbal reasoning. There was excellent internal consistency and good test-retest reliability. Tests of language comprehension and non-verbal reasoning correlated significantly with the C-BiLLT-Nor, indicating good construct validity. Factor analysis yielded a two-factor solution, suggesting it as a measure of receptive vocabulary, grammar, and overall language comprehension. No difference in results could be related to response mode, implying that gaze pointing is a viable option for children who cannot point with a finger. The C-BiLLT-Nor, with norms from 1;6-7;6 is a reliable measure of language comprehension.

PMID: 32706281

17. Evaluation of Risk Factors for Epilepsy in Pediatric Patients with Cerebral Palsy
Małgorzata Sadowska, Beata Sarecka-Hujar, Ilona Kopyta

Brain Sci. 2020 Jul 25;10(8):E481. doi: 10.3390/brainsci10080481.

Cerebral palsy (CP) is a set of etiologically diverse symptoms that change with the child's age. It is one of the most frequent causes of motor disability in children. CP occurs at a frequency of 1.5 to 3.0 per 1000 live-born children. CP often coexists with epilepsy, which is drug-resistant in a high number of cases. The aim of the present study was to analyze the associations between preconception, prenatal, perinatal, neonatal, and infancy risk factors for epilepsy in a group of pediatric patients with CP. We retrospectively analyzed 181 children with CP (aged 4-17 years at diagnosis), hospitalized at the Department of Pediatrics and Developmental Age Neurology in Katowice in the years 2008-2016. Division into particular types of CP was based on Ingram's classification. Data were analyzed using STATISTICA 13.0 (STATSOFT; Statistica, Tulsa, OK, USA). Epilepsy was diagnosed in 102 children (56.35%), of whom 44 (43%) had drug-resistant epilepsy; only in 15 cases (14.71%) was epilepsy susceptible to treatment. The incidence of epilepsy varied between the types of CP. It occurred significantly more often in children with tetraplegia (75%), ataxic form (83%), and mixed form (80%) in comparison to diplegia (32%) and hemiplegia (38%). Maternal hypertension was found to be a risk factor for epilepsy in CP patients (OR = 12.46, $p < 0.001$) as well as for drug-resistant epilepsy (the odds ratio (OR) = 9.86, $p = 0.040$). Delivery by cesarean section increased the risk of epilepsy in the CP patients over two-fold (OR = 2.17, $p = 0.012$). We observed also that neonatal convulsions significantly increased the risk for epilepsy (OR = 3.04, $p = 0.011$) as well as drug-resistant epilepsy (OR = 4.02, $p = 0.002$). In conclusion, maternal hypertension, neonatal convulsions, and delivery by cesarean section were the most important factors increasing the risk of epilepsy as well as drug-resistant epilepsy in the analyzed group of patients with CP.

PMID: 32722475

18. Cost-effectiveness of botulinum neurotoxin A versus surgery for drooling: a randomized clinical trial
Stijn Bekkers, Kim J van Ulsen, Eddy M M Adang, Arthur R T Scheffer, Frank J A van den Hoogen

Dev Med Child Neurol. 2020 Jul 24. doi: 10.1111/dmcn.14636. Online ahead of print.

Aim: This study compared the cost-effectiveness of botulinum neurotoxin A (BoNT-A) injections with two-duct ligation of the submandibular glands as treatment for severe drooling after one treatment cycle. Method: The study was part of a larger, partly single-blinded, randomized clinical trial (trialregister.nl identifier NTR3537). Data were collected between 2012 and 2017. Evaluation was at 32 weeks after one treatment cycle. Fifty-seven patients with cerebral palsy or other neurological, non-progressive disorders and severe drooling classified as having a drooling frequency ≥3 or a drooling severity ≥2, in whom conservative treatment was deemed ineffective, were randomized to treatment by BoNT-A or two-duct ligation. An incremental cost-effectiveness ratio (ICER) was calculated using the success rates as the measure of benefit. Treatment success was defined as a decrease ≥50% from baseline to 32 weeks in the subjective visual analogue scale for the severity of drooling or the objective drooling quotient. Results: Fifty-three patients were analysed (22 females, 31 males; mean age 11y, range 8-22y). Average costs for one treatment cycle, which included one BoNT-A injection, were €1929 (standard error 62) for BoNT-A and €3155 (standard error 99) for two-duct ligation. Treatment success was in favour of two-duct ligation (63% vs 27%; number needed to treat 3). The ICER was €34 per 1% gain in treatment success in favour of two-duct ligation versus BoNT-A. Interpretation: The additional cost of two-duct ligation is to some extent offset by a larger treatment success rate compared with BoNT-A. What this paper adds: Botulinum neurotoxin A (BoNT-A) is less expensive per percentage of success than two-duct ligation. The additional cost of two-duct ligation over BoNT-A is offset by greater treatment success.

PMID: 32706122

19. Pain in children with dyskinetic and mixed dyskinetic/spastic cerebral palsy
Clare T McKinnon, Prue E Morgan, Giuliana C Antolovich, Catherine H Clancy, Michael C Fahey, Adrienne R Harvey

Dev Med Child Neurol. 2020 Jul 25. doi: 10.1111/dmcn.14615. Online ahead of print.

Aim: To evaluate pain prevalence and characteristics in children and adolescents with predominant dyskinetic and mixed (dyskinetic/spastic) cerebral palsy (CP) motor types. Method: Seventy-five participants with a diagnosis of CP and confirmed dyskinetic or mixed (dyskinetic/spastic) motor type took part in a multisite cross-sectional study. The primary outcome was carer-reported pain prevalence (preceding 2wks) measured using the Health Utilities Index-3. Secondary outcomes were chronicity, intensity, body locations, quality of life, and activity impact. Results: Mean participant age was 10 years 11 months (SD 4y 2mo, range 5-18y). There were 44 males and 31 females and 37 (49%) had predominant dyskinetic CP. Pain was prevalent in 85% and it was chronic in 77% of participants. Fifty-two per cent experienced moderate-to-high carer-reported pain intensity, which was significantly associated with predominant dyskinetic motor types (p=0.008). Pain occurred at multiple body locations (5 out of 21), with significantly increased numbers of locations at higher Gross Motor Function Classification System levels (p=0.02). Face, jaw, and temple pain was significantly associated with predominant dyskinetic motor types (p=0.005). Poorer carer proxy-reported quality of life was detected in those with chronic pain compared to those without (p=0.03); however, chronic pain did not affect quality of life for self-reporting participants. Interpretation: Pain was highly prevalent in children and adolescents with predominant dyskinetic and mixed (dyskinetic/spastic) motor types, highlighting a population in need of lifespan pain management.

PMID: 32710570

20. Psychological morbidity among adults with cerebral palsy and spina bifida
Mark D Peterson, Paul Lin, Neil Kamdar, Elham Mahmoudi, Christina N Marsack-Topolewski, Heidi Haapala, Karin Muraszko, Edward A Hurvitz

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Background: Very little is known about the risk of developing psychological morbidities among adults living with cerebral palsy (CP) or spina bifida (SB). The objective of this study was to compare the incidence of and adjusted hazards for psychological morbidities among adults with and without CP or SB. Methods: Privately insured beneficiaries were included if they had an International Classification of Diseases, Ninth revision, Clinical Modification diagnostic code for CP or SB (n = 15 302). Adults without CP or SB were also included (n = 1 935 480). Incidence estimates of common psychological morbidities were compared at 4-years of enrollment. Survival models were used to quantify unadjusted and adjusted hazard ratios for incident psychological morbidities. Results: Adults living with CP or SB had a higher 4-year incidence of any psychological morbidity (38.8% v. 24.2%) as compared to adults without CP or SB, and differences were to a clinically meaningful extent. Fully adjusted survival models demonstrated that adults with CP or SB had a greater hazard for any psychological morbidity [hazard ratio (HR): 1.60; 95% CI 1.55-1.65], and all but one psychological disorder (alcohol-related disorders), and ranged from HR: 1.32 (1.23, 1.42) for substance disorders, to HR: 4.12 (3.24, 5.25) for impulse control disorders. Conclusions: Adults with CP or SB have a significantly higher incidence of and risk for common psychological morbidities, as compared to adults without CP or SB. Efforts are needed to facilitate the development of improved clinical screening algorithms and early interventions to reduce the risk of disease onset/progression in these higher-risk populations.

PMID: 32713401

21. Incidence of invasive Group B Streptococcal infection and the risk of infant death and cerebral palsy: a Norwegian Cohort Study
Maren Mynarek, Solveig Bjellmo, Stian Lydersen, Jan E Afset, Guro L Andersen, Torstein Vik

Pediatr Res. 2020 Jul 29. doi: 10.1038/s41390-020-1092-2. Online ahead of print.

Background: Group B Streptococcus (GBS) is the leading cause of invasive neonatal infection worldwide. In high-income countries mortality rates are 4-10%, and among survivors of GBS meningitis, 30-50% have neurodevelopmental impairments. We hypothesized that invasive GBS infection was associated with increased risk of infant mortality and cerebral palsy (CP). Methods: All children born alive in Norway during 1996-2012 were included. Data were collected from three national registers. Invasive GBS infection during infancy was categorized into early-onset disease (EOD), late-onset (LOD), and very late-onset disease (VLOD). Primary outcomes were infant mortality and CP. Results: Invasive GBS infection was diagnosed in 625 children (incidence: 0.62 per 1000 live births; 95% confidence interval (CI): 0.57-0.67). The incidence of EOD was 0.41 (0.37-0.45), of LOD 0.20 (0.17-0.23), and of VLOD 0.012 (0.007-0.021). The annual incidence of LOD increased slightly. Among infected infants, 44 (7%) died (odds ratio (OR): 24.5; 95% CI: 18.0-33.3 compared with the background population). Among survivors, 24 (4.1%) children were later diagnosed with CP, compared with 1887 (0.19%) in the background population (OR: 22.9; 95% CI: 15.1-34.5). Conclusion: Despite a relatively low incidence of invasive GBS infection in Norway, the risk of death and CP remains high. Improvements in prevention strategies are needed. Impact: During the first decade of the twenty-first century, invasive GBS disease in infancy is still associated with high mortality.Despite the overall low incidence of invasive GBS disease, the incidence of LOD increased during the study period.The finding that invasive GBS infection in the neonatal period or during infancy is associated with an excess risk of CP, comparable to the risk following moderate preterm birth and moderate low Apgar scores, adds to the existing literature.The results of this study emphasize the importance of adhering to guidelines and the need for better prevention strategies.

PMID: 32726797

22. Cerebral palsy and associated complex congenital nasolacrimal duct obstruction and pediatric acute dacryocystitis
Nandini Bothra, Mohammad Javed Ali

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Cerebral palsy (CP) is a congenital syndrome with systemic manifestations secondary to a non-progressive lesion of the immature brain. It is associated with numerous cerebral and non-cerebral malformations. The present report describes a 2-year-old baby with spastic CP, diplegic type, associated with congenital cardiac malformations and right eye complex congenital nasolacrimal duct obstruction (CNLDO) (bony nasolacrimal duct stenosis and buried probe) requiring endoscopic dacryocystorhinostomy and left eye simple CNLDO which resolved on probing the system. This report also lays emphasis on the need for examination of the lacrimal drainage system in all patients with CP, so as to treat them appropriately and reduce the morbidity.

PMID: 32718225

23. Disparity of child/parent-reported quality of life in cerebral palsy persists into adolescence
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Aim: To examine the evolution of child-parent discrepancy in reporting quality of life (QoL) between childhood and adolescence in children with cerebral palsy (CP) and to investigate potential factors associated with such a discrepancy. Method: We used data from the SPARCLE (Study of PARticipation of Children with CP Living in Europe) study, a population -based cohort study of children with CP, aged 8 to 12 years at baseline (in 2004-2005), in nine European centres, who were followed up at the age of 13 to 17 years. The KIDSCREEN-52 Quality of Life measure was used at baseline and follow-up; 354 child-parent dyads out of 500 eligible dyads were followed up (201 males, 153 females). We used intraclass correlation coefficients (ICCs) to examine agreement between parent proxy-reports and self-reported QoL. We used linear regression to examine factors associated with child-parent discrepancy in QoL reporting. Results: Agreement was low to moderate (ICC=0.16-0.48) in childhood and in adolescence across all QoL domains. In four domains (moods and emotions, self-perception, relationship with parents and home life, and social support and peers), the extent of the discrepancy increased significantly between childhood and adolescence. Parenting stress, child pain, and child behaviour problems influenced parent proxy-reports during both childhood and adolescence. Interpretation: The points of view of the child and their parents should be treated as complementary to obtain better knowledge regarding the QoL of children and adolescents with CP.

PMID: 32710687

24. Intrapartum Fetal Monitoring
James J Arnold, Breanna L Gawrys

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Continuous electronic fetal monitoring was developed to screen for signs of hypoxic-ischemic encephalopathy, cerebral palsy, and impending fetal death during labor. Because these events have a low prevalence, continuous electronic fetal monitoring has a false-positive rate of 99%. The widespread use of continuous electronic fetal monitoring has increased operative and cesarean delivery rates without improved neonatal outcomes, but its use is appropriate in high-risk labor. Structured intermittent auscultation is an underused form of fetal monitoring; when employed during low-risk labor, it can lower rates of operative and cesarean deliveries with neonatal outcomes similar to those of continuous electronic fetal monitoring. However, structured intermittent auscultation remains difficult to implement because of barriers in nurse staffing and physician oversight. The National Institute of Child Health and Human Development terminology is used when reviewing continuous electronic fetal monitoring and delineates fetal risk by three categories. Category I tracings reflect a lack of fetal acidosis and do not require intervention. Category II tracings are indeterminate, are present in the majority of laboring patients, and can encompass monitoring predictive of clinically normal to rapidly developing acidosis. Presence of moderate fetal heart rate variability and accelerations with absence of recurrent pathologic decelerations provides reassurance that acidosis is not present. Category II tracing abnormalities can be addressed by treating reversible causes and providing intrauterine resuscitation, which includes stopping uterine-stimulating agents, fetal scalp stimulation and/or maternal repositioning, intravenous fluids, or oxygen. Recurrent deep variable decelerations can be corrected with amnioinfusion. Category III tracings are highly concerning for fetal acidosis, and delivery should be expedited if immediate interventions do not improve the tracing.

PMID: 32735438

25. Maternal Δ-aminolevulinic acid dehydratase 1-2 genotype enhances fetal lead exposure and increases the susceptibility to the development of cerebral palsy
Omaima Ibrahim Abdel Hamid, Eman El-Sayed Hassan Khayal, Sabry Abdel Rahman Tolba, Eman Elshahat Orabi

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Limited epidemiologic studies questioned the association between pre- and postnatal lead exposure and the development of cerebral palsy (CP). Moreover, the genotypes of δ-aminolevulinic acid dehydratase (δ-ALAD) in CP patients and their mothers and their association to the blood lead levels (BLLs) were not previously studied. This study aimed to evaluate the association between δ-ALAD gene polymorphism and BLL in cases of CP and their mothers. A case control study was carried out on 23 CP cases and equal number of healthy matched controls. The mothers of the included children were asked to answer a questionnaire involving the baseline clinical and demographic characteristics. Also, questionnaires were done to detect the sources of environmental lead exposure and screen lead exposure during the pregnancy period. BLL, δ-ALAD enzyme activity, and genetic analysis for ALAD G177C were done for each child and his mother. There was significant ($p < 0.001$) elevation of BLL in CP cases and their mothers that was positively correlated ($r = 0.436$, $p < 0.05$). There were progressive decreases in δ-ALAD activity with increasing BLL in both children and mothers ($p < 0.05$). There were non-significant ($p > 0.05$) differences between CP and the control group regarding frequency of ALAD G177C genotypes, while there was a significant ($p = 0.04$) increase in the frequency of ALAD 1-2 (GC) genotype in the mothers of the CP group associated with high BLL and significant decrease in δ-ALAD activity ($p < 0.001$). The study can indicate the significance of δ-ALAD gene polymorphism in the prenatal exposure to lead and the affection of the developing brain, pointing to the importance of controlling lead in pregnant women especially those with ALAD 1-2 genotype.

PMID: 32710353

26. School-age outcomes following intraventricular haemorrhage in infants born extremely preterm
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Objective: To determine the associations of different grades of intraventricular haemorrhage (IVH), particularly grades 1 and 2, with neurodevelopmental outcomes at 8 years of age in children born extremely preterm. Design: Population-based cohort study. Setting: State of Victoria, Australia. Patients: Survivors born at <28 weeks' gestational age (n=546) and matched term-born controls (n=679) from three distinct eras, namely, those born in 1991-1992, 1997 and 2005. Exposure: Worst grade of IVH detected on serial neonatal cranial ultrasound. Outcome measures: Intellectual ability, executive function, academic skills, cerebral palsy and motor function at 8 years. Results: There was a trend for increased motor dysfunction with increasing severity of all grades of IVH, from 24% with no IVH, rising to 92% with grade 4 IVH. Children with grade 1 or 2 IVH were at higher risk of developing cerebral palsy than those without IVH (OR 2.24, 95% CI 1.21 to 4.16). Increased rates of impairment in intellectual ability and academic skills were observed with higher grades of IVH, but not for grade 1 and 2 IVH. Parent-rated executive functioning was not related to IVH. Conclusion: While low-grade IVH is generally considered benign, it was associated with higher rates of cerebral palsy in school-aged children born EP, but not with intellectual ability, executive function, academic skills or overall motor function. Higher grades of IVH were associated with higher rates and risks of impairment in motor function, intellectual ability and some academic skills, but not parental ratings of executive function.

PMID: 32732377

27. Ventilation, oxidative stress and risk of brain injury in preterm newborn
Laura Cannavò, Immacolata Rulli, Raffaele Falsaperla, Giovanni Corsello, Eloisa Gitto

Review Ital J Pediatr. 2020 Jul 23;46(1):100. doi: 10.1186/s13052-020-00852-1.

Preterm infants have an increased risk of cognitive and behavioral deficits and cerebral palsy compared to term born babies. Especially before 32 weeks of gestation, infants may require respiratory support, but at the same time, ventilation is known to induce oxidative stress, increasing the risk of brain injury. Ventilation may cause brain damage through two pathways: localized cerebral inflammatory response and hemodynamic instability. During ventilation, the most important causes of pro-inflammatory cytokine release are oxygen toxicity, barotrauma and volutrauma. The purpose of this review was to analyze the mechanism of ventilation-induced lung injury (VILI) and the relationship between brain injury and VILI in order to provide the safest possible respiratory support to a premature baby. As gentle ventilation from the delivery room is needed to reduce VILI, it is recommended to start ventilation with 21-30% oxygen, prefer a non-invasive respiratory approach and, if mechanical ventilation is required, prefer low Positive End-Expiratory Pressure and tidal volume.

PMID: 32703261