Interventions and Management

1. Perinatal Brain Injury: Mechanisms, Prevention, and Outcomes.
Novak CM, Ozen M, Burd I.

Perinatal brain injury may lead to long-term morbidity and neurodevelopmental impairment. Improvements in perinatal care have resulted in the survival of more infants with perinatal brain injury. The effects of hypoxia-ischemia, inflammation, and infection during critical periods of development can lead to a common pathway of perinatal brain injury marked by neuronal excitotoxicity, cellular apoptosis, and microglial activation. Various interventions can prevent or improve the outcomes of different types of perinatal brain injury. The objective of this article is to review the mechanisms of perinatal brain injury, approaches to prevention, and outcomes among children with perinatal brain injury.

PMID: 29747893

Hoare B, Ditchfield M, Thorley M, Wallen M, Bracken J, Harvey A, Elliott C, Novak I, Crichton A.

BACKGROUND: Motor outcomes of children with unilateral cerebral palsy are clearly documented and well understood, yet few studies describe the cognitive functioning in this population, and the associations between the two is poorly understood. Using two hands together in daily life involves complex motor and cognitive processes. Impairment in either domain may contribute to difficulties with bimanual performance. Research is yet to derive whether, and how, cognition affects a child's ability to use their two hands to perform bimanual tasks. METHODS/DESIGN: This study will use a prospective, cross-sectional multi-centre observational design. Children (aged 6-12 years) with unilateral cerebral palsy will be recruited from one of five Australian treatment centres. We will examine associations between cognition, bimanual performance and brain neuropathology (lesion type and severity) in a sample of 131 children. The primary outcomes are: Motor - the Assisting Hand Assessment; Cognitive - Executive Function; and Brain - lesion location on structural MRI. Secondary data collected will include: Motor - Box and Blocks, ABILHAND- Kids, Sword Test; Cognitive - standard neuropsychological measures of intelligence. We will use generalized linear modelling and structural equation modelling techniques to investigate relationships between bimanual performance, executive function and brain lesion location. DISCUSSION: This large multi-centre study will examine how cognition affects bimanual performance in children with unilateral cerebral palsy. First, it is anticipated that distinct relationships between bimanual performance and cognition (executive function) will be identified. Second, it is anticipated that interrelationships between bimanual performance and cognition will be associated with common underlying neuropathology. Findings have the potential to improve the specificity of existing upper limb interventions by providing more targeted treatments and influence the development of novel methods to improve both cognitive and motor outcomes in children with unilateral cerebral palsy.

PMID: 29739443
3. Bimanual tasks in unilateral cerebral palsy: one hand clapping?
Basu AP.
PMID: 29732543

Beretta E, Cesareo A, Biffi E, Schafer C, Galbiati S, Strazzer S.
Acquired brain injuries (ABIs) can lead to a wide range of impairments, including weakness or paralysis on one side of the body known as hemiplegia. In hemiplegic patients, the rehabilitation of the upper limb skills is crucial, because the recovery has an immediate impact on patient quality of life. For this reason, several treatments were developed to flank physical therapy (PT) and improve functional recovery of the upper limbs. Among them, Constraint-Induced Movement Therapy (CIMT) and robot-aided therapy have shown interesting potentialities in the rehabilitation of the hemiplegic upper limb. Nevertheless, there is a lack of quantitative evaluations of effectiveness in a standard clinical setting, especially in children, as well as a lack of direct comparative studies between these therapeutic techniques. In this study, a group of 18 children and adolescents with hemiplegia was enrolled and underwent intensive rehabilitation treatment including PT and CIMT or Armeo®Spring therapy. The effects of the treatments were assessed using clinical functional scales and upper limb kinematic analysis during horizontal and vertical motor tasks. Results showed CIMT to be the most effective in terms of improved functional scales, while PT seemed to be the most significant in terms of kinematic variations. Specifically, PT resulted to have positive influence on distal movements while CIMT conveyed more changes in the proximal kinematics. Armeo treatment delivered improvements mainly in the vertical motor task, showing trends of progresses of the movement efficiency and reduction of compensatory movements of the shoulder with respect to other treatments. Therefore, every treatment gave advantages in a specific and different upper limb district. Therefore, results of this preliminary study may be of help to define the best rehabilitation treatment for each patient, depending on the goal, and may thus support clinical decision.
PMID: 29732047

5. Cervical Myelopathy in a Child: A Rare Cause of Hypoventilation Syndrome Presenting with Type 2 Respiratory Failure.
Mondal A, Giri PP.
Hypoventilation syndrome leading to Type 2 respiratory failure is not a rare cause of Pediatric Intensive Care Unit admission and mechanical ventilation. Common causes in pediatric population are Guillain-Barre syndrome and various central nervous system disorders such as encephalitis, traumatic brain injury, and drugs. Any injury or disease in the cervical cord can also produce respiratory paralysis causing respiratory failure. Here, we present two cases of mixed cerebral palsy with cervical myelopathy due to compression effect of fractured segments of first and second cervical vertebrae. Both of them presented with Type 2 respiratory failure.
PMID: 29743771

Dallas J, Sborov KD, Guidry BS, Chotai S, Bonfield CM.
OBJECTIVE Many patients undergoing spinal fusion for neuromuscular scoliosis have preexisting neurosurgical implants, including ventricular shunts (VSs) for hydrocephalus and baclofen pumps (BPs) for spastic cerebral palsy. Recent studies have discussed a possible increase in implant complication rates following spinal fusion, but published data are inconclusive. The authors therefore, sought to investigate: 1) the rate of implant complications following fusion, 2) possible causes of these complications, and 3) factors that place patients at higher risk for implant-related complications. METHODS Cases involving pediatric patients with a preexisting VS or BP who underwent spinal fusion for scoliosis correction between 2005 and 2016 at a single tertiary children's hospital were retrospectively analyzed. Patient demographics, implant characteristics, spinal fusion details, neurosurgical follow-up, and implant complications in the 180 days following fusion were recorded and analyzed. RESULTS Overall, 75 patients who underwent scoliosis correction had preexisting implants: 39 had BPs, 31 VSs, and 5 both. The patients' mean age at fusion was 13.49 ± 2.78 years (range 3.62-18.81 years), and the mean time from the most recent
previous implant surgery to fusion was 5.70 ± 4.65 years (range 0.10-17.3 years). The mean preoperative and postoperative Cobb angles were 62.4° ± 18.9° degrees (range 20.9°-109.0°) and 23.5° ± 13.3° degrees (range 2.00°-67.3°), respectively. No VS complications were identified. Two patients with BPs were found to have complications (unintentional cutting of their BP catheter during posterior spinal fusion) within 180 days postfusion. There were no recorded neurosurgical implant infections, failures, fractures, or dislodgements. Although 10 patients required at least 1 surgical procedure for irrigation and debridement of the spine wound following fusion, there were no abdominal or cranial implant wound infections requiring revision, and no implants required removal. CONCLUSIONS The results of this study suggest that spinal fusion for scoliosis correction does not increase the rates of complications involving previously placed neurosurgical implants. A large-scale, prospective, multicenter study is needed to fully explore and confirm this finding.

PMID: 29726795


BACKGROUND: Early detection of changes at the muscular level before a contracture develops is important to gain knowledge about the development of deformities in individuals with spasticity. However, little information is available about muscle morphology in children with spastic diplegic cerebral palsy (CP) without contracture or equinus gait. Therefore, the aim of this study was to compare the gastrocnemius medialis (GM) and Achilles tendon architecture of children and adolescents with spastic CP without contracture or equinus gait to that of typically developing (TD) children. METHODS: Two-dimensional ultrasonography was used to assess the morphological properties of the GM muscle and Achilles tendon in 10 children with spastic diplegic CP (Gross Motor Function Classification System level I-II) and 12 TD children (mean age 12.0 (2.8) and 11.3 (2.5) years, respectively). The children with CP were not restricted in the performance of daily tasks, and therefore had a high functional capacity. Mean muscle and tendon parameters were statistically compared (independent t-tests or Mann-Whitney U-tests). RESULTS: When normalized to lower leg length, muscle-tendon unit length and GM muscle belly length were found to be significantly shorter (p < 0.05, effect size (ES) = 1.00 and 0.98, respectively) in the children with spastic CP. Furthermore, there was a tendency for increased Achilles tendon length when expressed as a percentage of muscle-tendon unit length (p = 0.08, ES = 0.80) in the individuals with CP. This group also showed shorter muscle fascicles (3.4 cm vs. 4.4 cm, p < 0.01, ES = 1.12) and increased fascicle pennation angle (21.9° vs. 18.1°, p < 0.01, ES = 1.36, respectively). However, muscle thickness and Achilles tendon cross-sectional area did not differ between groups. Resting ankle joint angle was significantly more plantar flexed (-26.2° vs. -20.8°, p < 0.05, ES = 1.06) in the children with CP. CONCLUSIONS: Morphological alterations of the plantar flexor muscle-tendon unit are also present in children and adolescents with mild forms of spastic CP. These alterations may contribute to functional deficits such as muscle weakness, and therefore have to be considered in the clinical decision-making process, as well as in the selection of therapeutic interventions.

PMID: 29743109


STUDY DESIGN: Retrospective, chart review. OBJECTIVES: The identification and management of pelvic obliquity in neuromuscular scoliosis can be difficult; therefore, appropriate evaluation of this pathology is important. Variations in presentation have resulted in various methods of measurement, without a consensus or gold standard measurement. This study is the evaluation of reliability of five methods commonly used to determine pelvic obliquity in the frontal plane. SUMMARY OF BACKGROUND DATA: Previous studies have used five different methods (ie, the Maloney method, the O'Brien method, the Osebold method, the Allen and Ferguson method, and the Lindseth method) to determine pelvic obliquity in the frontal plane. METHODS: Radiographic images of 50 patients with neuromuscular scoliosis involving the pelvic girdle were identified and evaluated by a team of 5 raters. Each rater was instructed to apply five commonly used methods of measuring pelvic obliquity. The same raters were asked to rate the same radiographs again after a minimum of one month. The interobserver reliability was assessed using the intraclass correlation coefficient (ICC) and the intraobserver analysis was assessed using the Pearson correlation. A rating greater than 0.8 was considered excellent reliability. RESULTS: The Maloney method showing the highest interobserver reliability of ICC of 0.965 and 0.964. The Lindseth method had the lowest ICC, but all the methods were considered highly reliable by the rating classification system. The Maloney method also showed the highest intraobserver reliability, ranging from 0.845 to 0.962. The Allen and Ferguson method had the lowest intraobserver reliability. CONCLUSIONS: Many of the methods tested had excellent reliability in this study, but our data suggest the Maloney method is the most reliable method of measuring pelvic obliquity on a frontal view radiograph. LEVEL OF EVIDENCE: Level III, retrospective, comparative study.

PMID: 29735134
Niklasch M, Boyer ER, Novacheck T, Dreher T, Schwartz M.

AIM: Femoral derotation osteotomy (FDO) can be conducted either proximally or distally to correct internal rotation gait (IRG) and increased anteversion in children with cerebral palsy (CP). Previous studies with limited numbers of participants have presented comparable short-term static and kinematic outcomes for both techniques. The objective of this retrospective multicentre study was to verify this thesis with a larger number of patients. METHOD: In total, 119 children with CP and IRG were included after matching the groups on preoperative mean stance hip rotation: 67 intertrochanteric (proximal group) FDO (average age at surgery 9y [SD 3y]); 52 supracondylar (distal group) FDO (average age at surgery 12y [SD 3y]). One random limb of each child was analyzed. Both transverse plane kinematic gait data and torsional parameters of clinical examination were assessed preoperatively and postoperatively. RESULTS: On average, both groups' mean hip rotation and midpoint of hip rotation improved postoperatively, and groups did not differ preoperatively or postoperatively for these variables (p>0.05). INTERPRETATION: The osteotomy location does not influence short-term gait kinematics nor static measurements. Therefore, the choice of performing proximal or distal FDO in children with CP for treatment of IRG and increased anteversion should be motivated by considerations other than these outcome parameters. What this paper adds Femoral derotation osteotomy (FDO) location does not influence mean stance hip rotation in cerebral palsy (CP). FDO location does not influence mid-point hip rotation in CP. FDO location should be motivated by concomitant procedures performed.

PMID: 29733439

10. [Detection study of walking segments of children with cerebral-palsy based on surface electromyographic signals].
Lou Z, Yao B, Yang J.

In this study, surface electromyography (sEMG) of the lower limbs of cerebral-palsy (CP) subjects in gait cycle was recorded and its parameters of gait cycle characters were analyzed to assess their clinical severity. Three algorithms, including integrated profile (IP), sample-entropy (SampEN) and smooth nonlinear energy operator (SNEO) algorithm, were applied to calculate the duration of walking sEMG segments in simulated SEMG signals. After that, the efficiency and accuracy were compared among these three algorithms. SNEO was then selected as the optimal algorithm among the three algorithms and employed for real sEMG signal processing of CP subjects. The results indicated that there was no significant difference in the accuracy of sEMG segment detection for the three algorithms. However, the computation speed of SNEO algorithm was much faster than those of the others and thus it was a suitable algorithm for detecting walking sEMG segments of CP subjects. In addition, the positive correlation was found between the clinical severity and the mean duration of walking sEMG segments in CP subjects. The results indicated that there was a significant difference in the three groups of CP subjects with different levels of severity. Our findings showed that the mean duration of walking sEMG segments could be considered as an assistant index to evaluate the clinical severity of CP subjects.

PMID: 29745498


Background: Patients with cerebral palsy (CP) typically receive limited physical therapy services. However, the Nintendo Wii system offers a simple and affordable mode of virtual reality therapy. There are no clinical trials assessing the Nintendo Wii balance board for improving standing balance in CP. METHODS: This randomised clinical trial will evaluate the effectiveness of an 18-session/six-week protocol using Wii therapy (W-t) compared with conventional therapy (C-t) in Chilean CP patients. The C-t group will perform the typical exercises prescribed by physical therapists for 40 min each session. W-t will consist of a virtual reality training session using the Nintendo Wii balance board console for 30 min each session. The primary outcome variable is the area of centre-of-pressure (CoP) sway (CoPSway). The secondary outcomes are the standard deviation (SDML; SDAP) and velocity (VML; VAP) of CoP in the ML and AP directions. For a mean difference of 21.5 cm² (CoPSway) between the groups, we required a minimum of 16 participants in each group. Data will be collected at baseline (week 0), during the study (weeks 2 and 4), at the end of the study (week 6), and during the follow-up (weeks 8 and 10). Measurements of postural control during quiet standing for both groups will be assessed on a force platform AMTI OR67. Discussion: This is the first trial that measures and compares the effects of a Nintendo Wii Balance Board exercise programme on standing balance.
in children with cerebral palsy compared to conventional therapy.

PMID: 29740634

Stevenson RD.

PMID: 29740808

Chiarello LA, Bartlett DJ, Palisano RJ, McCoy SW, Jeffries L, Fiss AL, Wilk P.

PURPOSE: To identify child, family, and service determinants of playfulness of young children with cerebral palsy.
METHODS: Participants were 429 children, 18-60 months. Children were divided into two groups, Gross Motor Function Classification System levels I-II and III-V. Therapists collected data on body functions and gross motor function; parents provided information about children's health conditions and adaptive behavior, family life, and services. One year after the beginning of the study, therapists assessed children's playfulness. Data were analyzed using structural equation modeling.
RESULTS: Higher gross motor function was associated with higher playfulness for both groups. Greater impact of health conditions on daily life was associated with lower playfulness for children in levels I-II. More effective adaptive behavior was associated with higher playfulness, and higher parent perception of therapists' family-centeredness was associated with lower playfulness for children in levels III-V. CONCLUSION: Supporting gross motor function, health, and adaptive behavior may foster playfulness.

PMID: 29746800

Molyneux E.

PMID: 29736991

15. Language development in deaf or hard-of-hearing children with additional disabilities: type matters!

BACKGROUND: This study examined language development in young children with hearing loss and different types of additional disabilities (ADs). METHOD: A population-based cohort of 67 children who were enrolled in the Longitudinal Outcomes of Children with Hearing Impairment study took part. Language ability was directly assessed at 3 and 5 years of age using the Preschool Language Scale, Fourth Edition and the Peabody Picture Vocabulary Test, Fourth Edition. Standard scores were used to enable comparison with age-based expectations for typically developing children. RESULTS: Analysis of variance showed that, across the total cohort, children's language scores remained stable over the 2-year period. However, this overall stability masked a significant difference between children with different types of ADs; in particular, children with autism, cerebral palsy and/or developmental delay showed a decline in standard scores, whereas children with other disabilities showed a relative improvement. In addition, larger improvements in receptive vocabulary were associated with use of oral communication only. CONCLUSIONS: The results suggest that type of AD can be used to gauge expected language development in the population of children with hearing loss and ADs when formal assessment of cognitive ability is not feasible.

PMID: 29732729

Stasolla F, Caffo AO, Perilli V, Boccasini A, Damiani R, D'Amico F.
OBJECTIVES: To extend the use of assistive technology for promoting adaptive skills of children with cerebral palsy. To assess its effects on positive participation of ten participants involved. To carry out a social validation recruiting parents, physiotherapists and support teachers as external raters. METHOD: A multiple probe design was implemented for Studies I and II. Study I involved five participants exposed to a combined program aimed at enhancing choice process of preferred items and locomotion fluency. Study II involved five further children for a combined intervention finalized at ensuring them with literacy access and ambulation responses. Study III recruited 60 external raters for a social validation assessment. RESULTS: All participants improved their performance, although differences among children occurred. Indices of positive participation increased as well. Social raters favorably scored the use of both technology and programs. CONCLUSION: Assistive technology-based programs were effective for promoting independence of children with cerebral palsy. Implications for Rehabilitation: A basic form of assistive technology such as a microswitch-based program may be useful and helpful for supporting adaptive skills of children with cerebral palsy and different levels of functioning. The same program may improve the participants’ indices of positive participation and constructive engagement with beneficial effects on their quality of life. The positive social rating provided by external experts sensitive to the matter may recommend a favorable acceptance and implementation of the program in daily settings.

PMID: 29732901

Palisano RJ, Avery L, Gorter JW, Galuppi B, McCoy SW.

AIM: To determine the stability of the Gross Motor Function Classification System (GMFCS), Manual Ability Classification System (MACS), and Communication Function Classification System (CFCS) over 1-year and 2-year intervals using a process for consensus classification between parents and therapists. METHOD: Participants were 664 children with cerebral palsy (CP), 18 months to 12 years of age, one of their parents, and 90 therapists. Consensus between parents and therapists on level of function was ≥92% for the GMFCS, MACS, and CFCS. A linearly weighted kappa coefficient of ≥0.75 was the criterion for stability. RESULTS: Kappa coefficients varied from 0.76 to 0.88 for the GMFCS, 0.59 to 0.73 for the MACS, and 0.57 to 0.77 for the CFCS. For children younger than 4 years of age, level of function did not change for 58.2% on the GMFCS, 30.3% on the MACS, and 39.3% on the CFCS. For children 4 years of age or older, level of function did not change for 72.3% on the GMFCS, 49.1% on the MACS, and 55% on the CFCS. INTERPRETATION: The findings support repeated classification of children over time. The kappa coefficients for the GMFCS are attributed to descriptions of levels for each age band. Consensus classification facilitates discussion between parents and professionals that has implications for shared decision-making. WHAT THIS PAPER ADDS: The findings support repeated classification of children over time. Stability was higher for the Gross Motor Function Classification System than the Manual Ability Classification System and Communication Function Classification System. The function of younger children was more likely to be reclassified. Percentage agreement between parents and therapists using consensus classification varied from 92% to 97%. The intraclass correlation coefficient overestimated stability compared with the weighted kappa coefficient.

PMID: 29726578

18. The reliability and validity of the Korean version of the Fullerton Advanced Balance scale in children with cerebral palsy.
Sim YJ MSc, PT, Kim GM PhD, PT, Yi CH PhD, PT.

The Fullerton Advanced Balance (FAB) scale is a multi-item balance assessment test designed to measure balance in relatively higher functioning individuals. The purpose of this study was to examine the reliability and validity of the Korean version of the FAB in children with cerebral palsy (CP). A total of 40 children with CP participated in this study. The internal consistency of the FAB was performed using the Cronbach alpha coefficient and the test-retest reliability was assessed. To verify the concurrent validity, scores on the FAB were compared with the pediatric balance scale (PBS) using the Spearman correlation coefficient. In addition, exploratory factor analysis was measured to explore the construct validity. The FAB showed satisfactory internal consistency (Cronbach’s alpha value = 0.92) and excellent test-retest reliability (ICC = 0.99). Concurrent validity was positively correlated with the FAB and PBS (r = 0.60, p < 0.001). Exploratory factor analysis revealed two dominant factors that explained 69.85% of the total variance of the scale. The FAB is a reliable and valid tool that can be used to measure the balance abilities in children with CP.

PMID: 29733748
19. Prolonged length of stay for acute hospital admissions as the increasing of age: A nationwide population study for Taiwan's patients with cerebral palsy.

Chiang KL, Huang CY, Fan HC, Kuo FC.


BACKGROUND: Studies investigating reasons for the admission and the associated lengths of stay (LOSs) among cerebral palsy (CP) patients are limited. This study determined common reasons for acute hospitalizations and the LOSs among children, adolescents, and young adults with CP. METHODS: We performed a secondary analysis of data. CP patients aged 4-32.9 years were identified by CP registry in the catastrophic illness patient registry of the 2010 Taiwan National Health Insurance Research Database. Data of admission claims from 2010 to 2011 were analyzed. Reasons for admissions were identified according to International Classification of Diseases codes. Common reasons, frequencies of admissions for each reason, and LOSs were reported. RESULTS: Pneumonia, other respiratory problems, and epilepsy were the top three reasons for admissions in all groups. Other common reasons in all groups were sepsis, other respiratory infections, and gastrointestinal problems. The reasons specific to children included orthopedic issues; ear, nose, and throat problems; and urinary tract infections (UTIs). In youths, scoliosis, and contractures, were unique reasons. In young adults, UTIs, blood problems, and mental illness, were special reasons. Most admission reasons appeared to prolong LOS, and the LOS exhibited an increasing trend as age increased. CONCLUSION: The results implied that patients with CP are more susceptible to most disease invasions. Our results also suggest that the current care system in Taiwan is unsuitable for patients with CP. These results can be used as guidance for planning effective multidisciplinary assessments in the future.

PMID: 29739652


Whitney DG, Hurvitz EA, Ryan JM, Devlin MJ, Caird MS, French ZP, Ellenberg EC, Peterson MD.


Purpose: Individuals with cerebral palsy (CP) are at increased risk for frailty and chronic disease due to factors experienced throughout the lifespan, such as excessive sedentary behaviors and malnutrition. However, little is known about noncommunicable diseases (NCDs) and multimorbidity profiles in young adults with CP. The study objective was to compare NCD and multimorbidity profiles between young adults with and without CP. Methods: A clinic-based sample of adults (18-30 years) with (n=452) and without (n=448) CP was examined at the University of Michigan Medical Center. The prevalence and predictors of 13 NCDs were evaluated, including existing diagnoses or historical record of musculoskeletal, cardiometabolic, and pulmonary morbidities. The level of motor impairment was determined by the Gross Motor Function Classification System (GMFCS) and stratified by less vs more severe motor impairment (GMFCS I-III vs IV-V). Logistic regression was used to determine the odds of NCD morbidity and multimorbidity in adults with CP compared to adults without CP, and for GMFCS IV-V compared to GMFCS I-III in those with CP, after adjusting for age, sex, body mass index, and smoking. Results: Adults with CP had a higher prevalence of osteopenia, osteoporosis, hypertension, myocardial infarction, hyperlipidemia, asthma, and multimorbidity compared to adults without CP, and higher odds of musculoskeletal (odds ratio [OR]: 6.97) and cardiometabolic morbidity (OR: 1.98), and multimorbidity (OR: 2.67). Adults with CP with GMFCS levels IV-V had a higher prevalence of osteopenia/osteoporosis, osteoarthritis, hypertension, other cardiovascular conditions, pulmonary embolism, and multimorbidity, and higher odds of musculoskeletal (OR: 3.41), cardiometabolic (OR: 2.05), pulmonary morbidity (OR: 1.42), and multimorbidity (OR: 3.45) compared to GMFCS I-III. Conclusion: Young adults with CP have a higher prevalence of chronic NCDs and multimorbidity compared to young adults without CP, which is pronounced in those with more severe motor impairment. These findings reiterate the importance of early screening for prevention of NCDs in CP.

PMID: 29750055


Polack S, Adams M, O'banion D, Baltussen M, Asante S, Kerac M, Gladstone M, Zuurmond M.


AIM: To assess feeding difficulties and nutritional status among children with cerebral palsy (CP) in Ghana, and whether severity of feeding difficulties and malnutrition are independently associated with caregiver quality of life (QoL). METHOD: This cross-sectional survey included 76 children with CP (18mo-12y) from four regions of Ghana. Severity of CP was classified using the Gross Motor Function Classification System and anthropometric measures were taken. Caregivers rated
their QoL (using the Pediatric Quality of Life Inventory Family Impact Module) and difficulties with eight aspects of child feeding. Logistic regression analysis explored factors (socio-economic characteristics, severity of CP, and feeding difficulties) associated with being underweight. Linear regression was undertaken to assess the relationship between caregiver QoL and child malnutrition and feeding difficulties. RESULTS: underweight, 54% as stunted, and 58% as wasted. Reported difficulties with child's feeding were common and were associated with the child being underweight (odds ratio 10.7, 95% confidence interval 2.3-49.6) and poorer caregiver QoL (p<0.001). No association between caregiver QoL and nutritional status was evident. INTERPRETATION: Among rural, low resource populations in Ghana, there is a need for appropriate, accessible caregiver training and support around feeding practices of children with CP, to improve child nutritional status and caregiver well-being. What this paper adds Malnutrition is very common among children with cerebral palsy in this rural population in Ghana. Feeding difficulties in this population were strongly associated with being underweight. Feeding difficulties were associated with poorer caregiver quality of life (QoL). Child nutritional status was not associated with caregiver QoL.

PMID: 29736993

22. Do preterm infants with a birth weight ≤1250 g born to single-parent families have poorer neurodevelopmental outcomes at age 3 than those born to two-parent families?


OBJECTIVE: Investigate neurodevelopmental outcomes at 3 years corrected age in infants with a birth weight ≤1250 g born to single parents. STUDY DESIGN: Infants born between 1995 and 2010 with a birth weight ≤1250 g were considered eligible. Primary outcome was neurodevelopmental impairment; considered present if a child had any of the following: cerebral palsy, cognitive delay, visual impairment, or deafness/neurosensoric hearing impairment. Univariate and multivariate analyses were performed. RESULT: A total of 1900 infants were eligible for inclusion. Follow-up data were available for 1395; 88 were born to a single parent. Infants in the single-parent group had higher mortality (18% vs. 11%, p = 0.009), IQ ≥1 SD below the mean (40% vs. 21%, p = 0.001) and any neurodevelopmental impairment (47% vs. 29%, p = 0.003). Single-parent family status, maternal education, bronchopulmonary dysplasia and severe neurological injury were significant predictors of intellectual impairment at 3 years corrected age. CONCLUSION: Preterm infants with a birth weight ≤1250 g born to single parents at birth have poorer intellectual functioning at 3 years corrected age.

PMID: 29740187

23. Perinatal Outcome and Long-Term Neurodevelopment after Intracranial Haemorrhage due to Fetal and Neonatal Alloimmune Thrombocytopenia.

Winkelhorst D, Kamphuis MM, Steggerda SJ, Rijken M, Oepkes D, Lopriore E, van Klink JMM.


OBJECTIVES: To evaluate the perinatal and long-term neurodevelopmental outcome in a cohort of children with intracranial haemorrhage (ICH) due to fetal and neonatal alloimmune thrombocytopenia (FNAIT) and to clearly outline the burden of this disease. SUBJECTS AND METHODS: We performed an observational cohort study and included all consecutive cases of ICH caused by FNAIT from 1993 to 2015 at Leiden University Medical Centre. Neurological, motor, and cognitive development were assessed at a minimum age of 1 year. The primary outcome was adverse outcome, defined as perinatal death or severe neurodevelopmental impairment (NDI). Severe NDI was defined as any of the following: cerebral palsy (Gross Motor Function Classification System [GMFCS] level ≥II), bilateral deafness, blindness, or severe motor and/or cognitive developmental delay (≤2 SD). RESULTS: In total, 21 cases of ICH due to FNAIT were included in the study. The perinatal mortality rate was 10/21 (48%). Long-term outcome was assessed in 10 children (n = 1 lost to follow-up). Severe and moderate NDI were diagnosed in 6/10 (60%) and 1/10 (10%) of the surviving children. The overall adverse outcome, including perinatal mortality or severe NDI, was 16/20 (80%). CONCLUSIONS: The risk of perinatal death or severe NDI in children with ICH due to FNAIT is high. Only screening and effective preventive treatment can avoid this burden.

PMID: 29730660


Vanzo RJ, Twede H, Ho KS, Prasad A, Martin MM, South ST, Wassman ER.


Copy number variants (CNV)s involving KANK1 are generally classified as variants of unknown significance. Several clinical case reports suggest that the loss of KANK1 on chromosome 9p24.3 has potential impact on neurodevelopment. These case
studies are inconsistent in terms of patient phenotype and suspected pattern of inheritance. Further complexities arise because these published reports utilize a variety of genetic testing platforms with varying resolution of the 9p region; this ultimately causes uncertainty about the impacted genomic coordinates and gene transcripts. Beyond these case reports, large case-control studies and publicly available databases statistically cast doubt as to whether variants of KANK1 are clinically significant. However, these large data sources are neither easily extracted nor uniformly applied to clinical interpretation. In this report we provide an updated analysis of the data on this locus and its potential clinical relevance. This is based on a review of the literature as well as 28 patients who harbor a single copy number variant involving KANK1 with or without DOCK8 (27 of whom are not published previously) identified by our clinical laboratory using an ultra-high resolution chromosomal microarray analysis. We note that 13 of 16 patients have a documented diagnosis of autism spectrum disorder (ASD) while only two, with documented perinatal complications, have a documented diagnosis of cerebral palsy (CP). A careful review of the CNVs suggests a transcript-specific effect. After evaluation of our case series and reconsideration of the literature, we propose that KANK1 aberrations do not frequently cause CP but cannot exclude that they represent a risk factor for ASD, especially when the coding region of the shorter, alternate KANK1 transcript (termed "transcript 4" in the UCSC Genome Browser) is impacted.

PMID: 29729439