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NEWS
Cerebral Palsy Research

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Interventions and Management

1. Res Dev Disabil. 2014 Oct 22;36C:162-174. doi: 10.1016/j.ridd.2014.09.011. [Epub ahead of print]

Evaluation and characterization of manual reaching in children with cerebral palsy: A systematic review.

Visicato LP1, da Costa CS2, Damasceno VA2, de Campos AC2, Rocha NA2.

Manual reaching is used daily to perform manipulative tasks and activities of daily routine. Children with cerebral palsy (CP) have limitations in this activity, with functional loss as a possible consequence. This review aimed to gather studies that evaluated and characterized manual reaching in children with CP, with the purpose of identifying the aspects analyzed, as well as review and discuss the results in the studies and its relationship to the children's level of functionality. 17 studies were selected for this systematic review from the search in electronic databases. The studies showed that children with CP show deficits in several spatio-temporal variables of reaching compared to typical children, such as longer time to perform the activity, higher peak velocity, lower index of curvature, and greater number of units of motion, which indicates lower smoothness and linearity of the movements of upper limbs. The performance is influenced by the level of motor impairment and various manipulations of the task. However, more studies are needed that help translating these results into treatment strategies that facilitate the performance of manual activities in children with CP.

PMID: 25462477 [PubMed - as supplied by publisher]

2. BMC Pediatr. 2014 Dec 5;14(1):292. [Epub ahead of print]

Intensive training of motor function and functional skills among young children with cerebral palsy: a systematic review and meta-analysis.

Tinderholt Myrhaug H, Ostensjø S, Larun L, Odgaard-Jensen J, Jahnsen R.

Background: Young children with cerebral palsy (CP) receive a variety of interventions to prevent and/or reduce activity limitations and participation restrictions. Some of these interventions are intensive, and it is a challenge to identify the optimal intensity. Therefore, the objective of this systematic review was to describe and categorise intensive motor function and functional skills training among young children with CP, to summarise the effects of these interventions, and to examine characteristics that may contribute to explain the variations in these effects.Methods: Ten databases were searched for controlled studies that included young children (mean age less

than seven years old) with CP and assessments of the effects of intensive motor function and functional skills training. The studies were critically assessed by the Risk of bias tool (RoB) and categorised for intensity and contexts of interventions. Standardised mean difference were computed for outcomes, and summarised descriptively or in meta-analyses. Results: Thirty-eight studies were included. Studies that targeted gross motor function were fewer, older and with lower frequency of training sessions over longer training periods than studies that targeted hand function. Home training was most common in studies on hand function and functional skills, and often increased the amount of training. The effects of constraint induced movement therapy (CIMT) on hand function and functional skills were summarised in six meta-analyses, which supported the existing evidence of CIMT. In a majority of the included studies, equal improvements were identified between intensive intervention and conventional therapy or between two different intensive interventions. Conclusion: sDifferent types of training, different intensities and different contexts between studies that targeted gross and fine motor function might explain some of the observed effect variations. Home training may increase the amount of training, but are less controllable. These factors may have contributed to the observed variations in the effectiveness of CIMT. Rigorous research on intensive gross motor training is needed.

Systematic review registration number CRD42013004023.

PMID: 25475608 [PubMed - as supplied by publisher]

3. J Hand Ther. 2014 Oct 6. pii: S0894-1130(14)00146-X. doi: 10.1016/j.jht.2014.09.007. [Epub ahead of print]

Effects of taping the hand in children with cerebral palsy.

Keklicek H1, Uygur F2, Yakut Y2.

BACKGROUND: Thumb in palm deformity restricts hand function by preventing somatosensory input in children with cerebral palsy who have spasticity in their hands. OBJECTIVES: To investigate the effects of thenar palmar tape application with and without pressure on upper extremity function in children with cerebral palsy. METHOD: 45 children were randomly assigned to one of the thenar taping groups either with or without pressure or to the control group. Nine hole peg test and nine parts puzzle test were used to measure upper extremity function. The two study groups were evaluated initially, with taping 20 min later and 20 min after taping was removed. The control group was evaluated initially, 20 min later and again after 20 min. RESULTS: Intragroup analyses showed that initially there was a difference in favor of the control group: number of pegs placed in the hole in 25 s ($p = 0.032$); number of puzzle parts placed in the hole in 25 s ($p = 0.028$). Following 20 min of application, there was no longer any difference between the groups ($p = 0.458$, $p = 0.286$ respectively). This was accepted as a manifestation of the effectiveness of taping. Intergroup analyses also showed a carry over effect 20 min after removing the tape only in the palmar pressure group ($p = 0.004$ and $p = 0.014$). Conclusion: It was concluded that taping can be an effective option for repositioning the thumb and improves upper extremity function by controlling the thumb in palm mechanically and enabling sensorial input by maintaining the correct hand position.

PMID: 25446521 [PubMed - as supplied by publisher]

4. Res Dev Disabil. 2014 Oct 15;36C:87-101. doi: 10.1016/j.ridd.2014.09.024. [Epub ahead of print]

Efficacy of upper limb strengthening in children with Cerebral Palsy: A critical review.

Rameckers EA1, Janssen-Potten YJ2, Essers IM3, Smeets RJ2.

OBJECTIVE: This review focuses on the effects of strengthening interventions of the upper limb in children with Cerebral Palsy (CP). The strengthening intervention studies were divided in two categories: those based on stand-alone strength training, and those on strength training combined with other interventions. DATA SOURCES AND EXTRACTION: A search in all relevant databases was performed. DATA SYNTHESIS: Six articles were included: three randomized controlled trials (RCTs), two clinical trial (CT) and one case study. Effect sizes of strength training on muscle strength and function of the upper limb were calculated. Conclusion: There are no coherent recommendations for strength training, based on these studies. The causes include too much variety of types of training, level of intensity and duration. All of the reported upper limb strength training studies found an increase in

muscle strength. In addition, the quality of these studies was not high. More RCTs on strength training according to the official strength training guidelines are necessary to assess the impact and potential of strength training of the upper limb to improve the daily activities and participation in children with CP.

PMID: 25462469 [PubMed - as supplied by publisher]

5. BMC Neurol. 2014 Nov 30;14(1):221. [Epub ahead of print]

Neglect-like characteristics of developmental disregard in children with cerebral palsy revealed by event related potentials.

Zielinski IM, Steenbergen B, Baas C, Aarts P, Jongsma M.

Background: Children with unilateral Cerebral Palsy (CP) often show diminished awareness of the remaining capacity of their affected upper limb. This phenomenon is known as Developmental Disregard (DD). DD has been explained by operant conditioning. Alternatively, DD can be described as a developmental delay resulting from a lack of use of the affected hand during crucial developmental periods. We hypothesize that this delay is associated with a general delay in executive functions (EF) related to motor behavior, also known as motor EFs. Methods: Twenty-four children with unilateral CP participated in this cross-sectional study, twelve of them diagnosed with DD. To test motor EFs, a modified go/nogo task was presented in which cues followed by go- or nogo-stimuli appeared at either the left or right side of a screen. Children had to press a button with the hand corresponding to the side of stimulus presentation. Apart from response accuracy, Event-Related Potentials (ERPs) extracted from the ongoing EEG were used to register covert cognitive processes. ERP N1, P2, N2, and P3 components elicited by cue-, go-, and nogo-stimuli were further analyzed to differentiate between different covert cognitive processes. Results: Children with DD made more errors. With respect to the ERPs, the P3 component to go-stimuli was enhanced in children with DD. This enhancement was related to age, such that younger children with DD showed stronger enhancements. In addition, in DD the N1 component to cue- and go-stimuli was decreased. Conclusion: The behavioral results show that children with DD experience difficulties when performing the task. The finding of an enhanced P3 component to go-stimuli suggests that these difficulties are due to increased mental effort preceding movement. As age in DD mediated this enhancement, it seems that this increased mental effort is related to a developmental delay. The additional finding of a decreased N1 component in DD furthermore suggests a general diminished visuo-spatial attention. This effect reveals that DD might be a neuropsychological phenomenon similar to post-stroke neglect syndrome that does not resolve during development. These findings suggest that therapies aimed at reducing neglect could be a promising addition to existing therapies for DD.

PMID: 25433482 [PubMed - as supplied by publisher] Free full text

6. Res Dev Disabil. 2014 Nov 24;37C:9-16. doi: 10.1016/j.ridd.2014.10.050. [Epub ahead of print]

Comparison of dosage of intensive upper limb therapy for children with unilateral cerebral palsy: How big should the therapy pill be?

Sakzewski L1, Provan K2, Ziviani J3, Boyd RN2.

This study aimed to compare efficacy of two dosages of modified constraint induced movement therapy (mCIMT) and bimanual therapy on upper limb and individualized outcomes for children with unilateral cerebral palsy. This secondary analysis included two separate randomized trials that compared equal doses (high or low) of mCIMT to bimanual therapy; Study 1 (full dose - 60h) n=64 and; Study 2 (half dose - 30h) n=18 for children aged five to 16 years with unilateral cerebral palsy. Outcomes for both studies included the Melbourne Assessment of Unilateral Upper Limb Function, Assisting Hand Assessment, Jebsen Taylor Test of Hand Function and Canadian Occupational Performance Measure which were administered at baseline, three and 26 weeks. Mixed linear modelling was used to compare between dose (e.g. "full dose" to "half dose" of either mCIMT or bimanual therapy) on outcomes at three and 26 weeks post-intervention. There were no significant differences between groups at baseline, however, on average the half dose mCIMT group was younger with better hand function compared to the other groups. The full compared to half dose mCIMT group achieved greater gains in bimanual performance at three weeks and dexterity and quality of movement at 26 weeks. There were no between group differences for

bimanual therapy doses. Half dose groups receiving either mCIMT or bimanual therapy did not make significant within group gains on any upper limb motor outcome, however gains in occupational performance were clinically meaningful. These results suggest that a half dose (30h) of either mCIMT or bimanual therapy may not be sufficient to impact upper limb outcomes, but made clinically meaningful gains in occupational performance for school aged children with UCP.

PMID: 25460215 [PubMed - as supplied by publisher]

7. Clin Biomech (Bristol, Avon). 2014 Nov 5. pii: S0268-0033(14)00254-X. doi: 10.1016/j.clinbiomech.2014.10.009. [Epub ahead of print]

Impact of muscle activation on ranges of motion during active elbow movement in children with spastic hemiplegic cerebral palsy.

Sarcher A1, Raison M2, Ballaz L3, Lemay M3, Leboeuf F4, Trudel K5, Mathieu PA6.

BACKGROUND: Children with spastic hemiplegic cerebral palsy are restricted in their daily activities due to limited active ranges of motion of their involved upper limb, specifically at the elbow. Their impaired muscles are frequently targeted by anti-spastic treatments that reduce muscle tone. But these treatments do not necessarily improve the limb function. There is a lack of comprehensive knowledge of the quantitative relations between muscle activation and joint active ranges of motion. Consequently, the objective of this study is to quantify the impact of muscle activation on the elbow active ranges of motion. Methods: During voluntary elbow pronation/supination and extension/flexion movements, kinematic and electromyographic measurements were collected from the involved upper limb of 15 children with spastic hemiplegic cerebral palsy (mean age=8.7years, standard deviation=2.2) and the dominant upper limb of 15 age-matched children who are typically developing. Representative indicators of the muscle activation, such as the muscle co-activation, were extracted from the electromyographic measurements. FINDINGS: Muscle co-activation in the involved upper limb accounted for 78% and 59% of the explained variance of the supination and extension limited active ranges of motion respectively. The agonist and antagonist muscle activations were both longer in the involved upper limb. INTERPRETATIONS: This study succeeded in quantifying the impact of longer antagonist muscle activation on decreased elbow active ranges of motion in children with spastic hemiplegic cerebral palsy. Longer agonist muscle activation suggests that strengthening agonist muscles could increase the extension and supination ranges of motion, which constitutes a perspective of future clinical studies.

PMID: 25467763 [PubMed - as supplied by publisher]

8. Res Dev Disabil. 2014 Nov 11;36C:413-418. doi: 10.1016/j.ridd.2014.10.037. [Epub ahead of print]

Effect of the angle of shoulder flexion on the reach trajectory of children with spastic cerebral palsy.

Furuya M1, Ohata K2, Izumi K2, Matsubayashi J1, Tominaga W1, Mitani A3.

Many children with cerebral palsy (CP) use a wheelchair during activities of daily living and often extend their hand upward and downward to reach objects in a seated position in a wheelchair. However, the effect of shoulder position on reaching movements of children with CP is not established. The purpose of this study was to determine the effect of the angle of shoulder flexion on the reach trajectory of children with spastic CP. Seven children with mild CP [Manual Ability Classification System (MACS) levels I-II], five children with severe CP (MACS levels III-V) and six typically developing (TD) children participated. We prepared the device to have a top board with variable tilting angle in order to reduce the effect of gravity imposing on reaching movements. By using this device, the subjects could extend their arm by sliding it on the board to push a target button. The reaching movements were performed with the more affected hand at three angles (60°, 90° and 120°) of shoulder flexion and captured using a camera motion analysis system. Subjects in the TD and mild CP groups reached the target at 60°, 90° and 120° of shoulder flexion. Subjects of the severe CP group reached the target at 60° and 90° of shoulder flexion, but two of the subjects could not reach the target at 120° of shoulder flexion. The TD and mild CP groups showed smooth and

almost straight trajectories at all three angles of shoulder flexion; however, the reach trajectory in the subjects with severe CP changed with the angle of shoulder flexion. A large angle of shoulder flexion induced great outward deviation in the trajectory. These findings suggest that the difficulty of the reaching task is changed depending on the shoulder joint angle in children with severe CP and that therapeutic interventions for children with severe CP should be provided in a manner appropriate for the shoulder joint angle.

PMID: 25462501 [PubMed - as supplied by publisher]

9. J Phys Ther Sci. 2014 Nov;26(11):1745-1747. Epub 2014 Nov 13.

A Reliability of the Prototype Trunk Training System for Sitting Balance.

Jeong J, Park DS, Lee H, Eun S.

Purpose: Cerebral palsy is a disorder that affects balance in the sitting position. Cerebral palsy patients need trunk muscle strengthening and balance training. In order to improve trunk control sensory-motor control training is carried out on an unstable surface. We have developed a Trunk Training System (TTS) that can provide visual feedback using a tilt sensor for balance training in the sitting position. Before using the TTS for training children with cerebral palsy experiments were conducted with healthy adult subjects and the TTS to gather basic data for its improvement. Subjects: The subjects were 11 healthy men (n=3) and women (n=8). Methods: Subjects trained at two levels (5°, 10°), in four different directions (anterior, posterior, left, right), three times each. TTS outcome indices (stability index, performance time) were measured. [Results] The stability index and performance time showed high correlation (-0.6<r<1). The measurements of the different task levels and directions showed high reliability (0.9<a). Conclusion: The TTS may be used to evaluate the range of motion and execution capabilities of sitting balance. Additional experiments will be needed to investigate the validity of the TTS measurements.

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10. Res Dev Disabil. 2014 Oct 15;36C:72-77. doi: 10.1016/j.ridd.2014.09.016. [Epub ahead of print]

Capacity of adolescents with cerebral palsy on paediatric balance scale and Berg balance scale.

Jantakat C1, Ramrit S2, Emasithi A3, Siritaratiwat W4.

The Berg balance scale (BBS) and the paediatric balance scale (PBS) are reliable tools for measuring balance ability. However, reports of BBS and PBS scores in adolescent cerebral palsy have been limited. The objectives of this study were to investigate functional balance capacities, as tested with the BBS and PBS in adolescents with cerebral palsy, to compare the total PBS and BBS scores between Gross Motor Function Classification System-Expanded and Revised (GMFCS-E&R) levels and to compare the static balance PBS and BBS scores within each GMFCS-E&R level. Fifty-eight school-aged adolescents with cerebral palsy between the ages of 12 and 18 years with GMFCS-E&R levels of I to IV were recruited. The Kruskal-Wallis test was utilized to compare the median scores for the PBS and BBS between the different GMFCS-E&R levels. Wilcoxon signed-rank tests were performed to examine the differences in the static balance scores between the PBS and the BBS within the same GMFCS-E&R levels. The results reveal that there were differences in the BBS and PBS scores among the four GMFCS-E&R levels. A significant difference was found between the BBS and PBS scores only among the patients with cerebral palsy and level III GMFCS-E&R. The BBS and PBS are valid and reliable tools for clinical examination and for distinguishing between levels of functional balance in adolescents with cerebral palsy.

PMID: 25462467 [PubMed - as supplied by publisher]

11. Clin Orthop Surg. 2014 Dec;6(4):426-31. doi: 10.4055/cios.2014.6.4.426. Epub 2014 Nov 10.

Anterior knee pain in patients with cerebral palsy.

Choi Y1, Lee SH2, Chung CY1, Park MS1, Lee KM1, Sung KH3, Won SH1, Lee IH1, Choi IH4, Cho TJ4, Yoo WJ4, Lee SY5.

BACKGROUND: The aim of this study was to identify the risk factors for anterior knee pain in patients with cerebral palsy. Methods: This prospective study investigated the risk factors for anterior knee pain in 127 ambulatory patients with spastic cerebral palsy in terms of walking pain, resting pain, and provocative pain. Demographic data analysis and physical examination for measuring the knee flexion contracture and unilateral and bilateral popliteal angles were performed. Patellar height was measured on radiographs, and patella alta was identified. The risk factors for anterior knee pain were analyzed using multivariate analysis with a generalized estimating equation. RESULTS: Seventy-seven patients were found to have patella alta based on the radiographic measurements (60.6%). Overall, sixteen patients (12.6%) had either unilateral or bilateral anterior knee pain. Of these, 6 patients showed a visual analogue scale (VAS) = 3, 9 patients showed 3 < VAS = 7, and one patient showed a VAS > 7. Age was found to be a significant risk factor for walking pain and resting pain with odds ratios (ORs) of 1.08 (95% confidence interval [CI], 1.02 to 1.14) and 1.09 (95% CI, 1.03 to 1.15), respectively. In the multivariate analysis, knee flexion contracture was a significant protective factor with an OR of 0.92 (95% CI, 0.85 to 0.98). Conclusion: Approximately 12.6% of ambulatory patients with spastic cerebral palsy were found to have anterior knee pain in our hospital-based cohort study. Age was found to be a significant risk factor for anterior knee pain while walking and resting.

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12. Arch Phys Med Rehabil. 2014 Oct 31. pii: S0003-9993(14)01209-X. doi: 10.1016/j.apmr.2014.09.039. [Epub ahead of print]

Biomechanical and clinical correlates of swing-phase knee flexion in individuals with spastic cerebral palsy who walk with flexed-knee gait.

Rha DW1, Cahill-Rowley K2, Young J3, Torburn L4, Stephenson K5, Rose J3.

OBJECTIVE: To identify clinical and biomechanical parameters that influence swing phase knee flexion and contribute to stiff-knee gait in individuals with spastic cerebral palsy (CP) and flexed-knee gait. DESIGN: Retrospective analysis of clinical data and gait kinematics collected from 2010-2013. SETTING: Motion & Gait Analysis Laboratory at Lucile Packard Children's Hospital at Stanford. PARTICIPANTS: Individuals with spastic CP (GMFCS I-III) who walked with flexed-knee gait = 20° at initial contact and had no prior surgery were included; the more-involved limb was analyzed. MAIN OUTCOME MEASURES: Magnitude and timing of peak knee flexion angle during swing (PKFSw) were analyzed with respect to clinical data, including passive range of motion and Selective Motor Control Assessment for the Lower Extremity (SCALE), and biomechanical data, including joint kinematics and hamstring, rectus femoris, and gastrocnemius muscle-tendon length during gait. RESULTS: Data from a total of 34 participants (20 males, 14 females; age 10.1±4.1, 5-20 years) demonstrated that achieving a higher magnitude of PKFSw correlated with higher maximum knee flexion velocity in swing (rho=0.582, p<0.001) and longer maximum length of rectus femoris (rho=0.491, p=0.003). In contrast, attaining earlier timing of PKFSw correlated with higher knee flexion velocity at toe-off (rho=-0.576, p<0.001), longer maximum length of gastrocnemius (rho=-0.355, p=0.039), and greater peak knee extension during single limb support phase (rho=-0.354, p=0.040). Conclusion: Results indicate that the magnitude and timing of PKFSw were independent and their biomechanical correlates differed, suggesting important treatment implications for both stiff-knee and flexed-knee gait.

PMID:25450128 [PubMed - as supplied by publisher]

13. Clin Biomech (Bristol, Avon). 2014 Nov 26. pii: S0268-0033(14)00290-3. doi: 10.1016/j.clinbiomech.2014.11.009. [Epub ahead of print]

Trunk movements during gait in cerebral palsy.

Attias M1, Bonnefoy-Mazure A2, Lempereur M3, Lascombes P4, De Coulon G4, Armand S5.

BACKGROUND: Lower limb deficits have been widely studied during gait in cerebral palsy, deficits in upper body have received little attention. The purpose of this research was to describe the characteristics of trunk movement of cerebral palsy children in terms of type of deficits (diplegia/hemiplegia) and gross motor function classification system (1, 2 or 3). Methods: Data from 92 cerebral palsy children, which corresponds to 141 clinical gait analysis, were retrospectively selected. Kinematic parameters of trunk were extracted from thorax and spine angles in the sagittal, transverse and coronal planes. The range of motion and the mean positions over the gait cycle were analysed. Intra-group differences between the children with diplegia or hemiplegia, gross motor function classification systems 1 to 3 and typically developing participants were analysed with Kruskal-Wallis tests and post hoc tests. Pearson correlation coefficients between the gait profile score normalised walking speed and kinematic parameters of the thorax were assessed. FINDINGS: The results revealed: 1) the range of motion of the thorax and spine exhibited more significant differences between groups than the mean positions; 2) greater levels of impairment were associated with higher thorax range of motion, and 3) the children with diplegia and gross motor function classification system 3 exhibited a greater range of motion for all planes with the exception of spine rotation. INTERPRETATION:
This study confirmed that greater levels of impairment in cerebral palsy are associated with greater thorax range of motion during gait. The thorax plays an important role during gait in cerebral palsy.

PMID: 25480360 [PubMed - as supplied by publisher]

14. Gait Posture. 2014 Nov 10. pii: S0966-6362(14)00753-X. doi: 10.1016/j.gaitpost.2014.10.027. [Epub ahead of print]

Kinematic foot types in youth with equinovarus secondary to hemiplegia.

Krzak JJ1, Corcos DM2, Damiano DL3, Graf A4, Hedeker D5, Smith PA6, Harris GF7.

BACKGROUND: Elevated kinematic variability of the foot and ankle segments exists during gait among individuals with equinovarus secondary to hemiplegic cerebral palsy (CP). Clinicians have previously addressed such variability by developing classification schemes to identify subgroups of individuals based on their kinematics. OBJECTIVE: To identify kinematic subgroups among youth with equinovarus secondary to CP using 3-dimensional multi-segment foot and ankle kinematics during locomotion as inputs for principal component analysis (PCA), and K-means cluster analysis. Methods: In a single assessment session, multi-segment foot and ankle kinematics using the Milwaukee Foot Model (MFM) were collected in 24 children/adolescents with equinovarus and 20 typically developing children/adolescents. RESULTS: PCA was used as a data reduction technique on 40 variables. K-means cluster analysis was performed on the first six principal components (PCs) which accounted for 92% of the variance of the dataset. The PCs described the location and plane of involvement in the foot and ankle. Five distinct kinematic subgroups were identified using K-means clustering. Participants with equinovarus presented with variable involvement ranging from primary hindfoot or forefoot deviations to deformtiy that included both segments in multiple planes. Conclusion: This study provides further evidence of the variability in foot characteristics associated with equinovarus secondary to hemiplegic CP. These findings would not have been detected using a single segment foot model. The identification of multiple kinematic subgroups with unique foot and ankle characteristics has the potential to improve treatment since similar patients within a subgroup are likely to benefit from the same intervention(s).

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15. J Phys Ther Sci. 2014 Nov;26(11):1667-1669. Epub 2014 Nov 13.

The Effect of Exercise Using a Sliding Rehabilitation Machine on the Gait Function of Children with Cerebral Palsy.

Lee YS1, Kim WB2, Park JW3.

Purpose: The purpose of the present study was to examine the effect of strength training using a sliding rehabilitation machine (SRM) on the gait function of cerebral palsy children. [Subjects and Methods: Thirteen children aged 6-18 years participated in the SRM training for 8 weeks (30 min/day, 2 times/week). The SRM is designed for the performance of a closed-kinetic chain exercise in which a tilt table is moved up and down using wheels on the table. Participants began in a position of flexion of the 3 lower joints (hips, knees, and ankles) on the SRM. In each exercise session, they extended and flexed the 3 joints. The level of exercise was set by changing the inclination of the tilt table. Functional gait ability was measured with the 6-minute walk test (6MWT), 10-m walk test (10MWT), and timed up-and-go test (TUG) before and after the training. Muscle strength was also measured using an isokinetic dynamometer. Results: Nine of the thirteen children completed the entire study. The peak torques of the knee extensor and flexor group muscles significantly improved after training with the SRM. The total distance of the 6 MWT significantly increased after training. The times of the 10 MWT and the TUG significantly improved after training. The changes in muscle tone were also investigated using the MAS (Modified Ashworth Scale) and Tardieu scale, but no significant changes were found in muscle tone between the pre- and post-test measurements. Conclusion: The findings demonstrate the effect of the SRM intervention which resulted in improved muscle strength and functional gait.

PMID: 25435673 [PubMed - as supplied by publisher] PMCID: PMC4242928 Free PMC Article

16. Res Dev Disabil. 2014 Nov 26;37C:95-101. doi: 10.1016/j.ridd.2014.10.053. [Epub ahead of print]

Motor imagery for walking: A comparison between cerebral palsy adolescents with hemiplegia and diplegia.

Molina M1, Kudlinski C2, Guilbert J2, Spruijt S3, Steenbergen B4, Jouen F5.

The goal of the study was to investigate whether motor imagery (MI) could be observed in cerebral palsy (CP) participants presenting a bilateral affected body side (diplegia) as it has been previously revealed in participants presenting a unilateral body affected sided (hemiplegia). MI capacity for walking was investigated in CP adolescents diagnosed with hemiplegia (n=10) or diplegia (n=10) and in adolescents with typical motor development (n=10). Participants were explicitly asked to imagine walking before and after actually walking toward a target located at 4m and 8m. Movement durations for executed and imagined trials were recorded. ANOVA and Pearson's correlation analyses revealed the existence of time invariance between executed and imagined movement durations for the control group and both groups of CP participants. However, results revealed that MI capacity in CP participants was observed for the short distance (4m) but not for the long distance (8m). Moreover, even for short distance, CP participants performed worse than typical adolescents. These results are discussed inline of recent researches suggesting that MI in CP participants may not depend on the side of the lesion.

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17. Phys Med Rehabil Clin N Am. 2015 Feb;26(1):57-67. doi: 10.1016/j.pmr.2014.09.005.

Pathophysiology of Muscle Contractures in Cerebral Palsy.

Mathewson MA1, Lieber RL2.

Patients with cerebral palsy present with a variety of adaptations to muscle structure and function. These pathophysiologic symptoms include functional deficits such as decreased force production and range of motion, in addition to changes in muscle structure such as decreased muscle belly size, increased sarcomere length, and altered extracellular matrix structure and composition. On a cellular level, patients with cerebral palsy have fewer

muscle stem cells, termed satellite cells, and altered gene expression. Understanding the nature of these changes may present opportunities for the development of new muscle treatment therapies.

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18. Hum Mov Sci. 2014 Nov 24;39C:65-72. doi: 10.1016/j.humov.2014.11.003. [Epub ahead of print]

Effects of whole body vibration on spasticity and lower extremity function in children with cerebral palsy.

Cheng HY1, Ju YY2, Chen CL3, Chuang LL4, Cheng CH5.

AIM: The aim of this study was to evaluate the effect of whole body vibration (WBV) on lower extremity spasticity and ambulatory function in children with cerebral palsy (CP) with a complete crossover design. METHOD: Sixteen participants aged 9.8(2.3) years received a 20-min WBV and a control condition in a counterbalanced order on two separate days. Change scores of each outcome variable were used to show the improvement. RESULTS: Repeated-measures analyses revealed significant differences in condition scores among variables including active range-of-motion (active ROM, increased), relaxation index (RI, increased), Modified Ashworth Scale (MAS, decreased), timed up-and-go (TUG, decreased), and Six Minute Walk Test (6MWT, increased). Significant differences were also found in time change scores for MAS and 6MWT. Correlation results revealed that TUG was significantly correlated with RI (r=-.512, p=.042), and 6MWT (r=-.700, p=.003). INTERPRETATION: This study suggested that WBV intervention can control the spasticity, enhance ambulatory performance and increase active ROM. Along with previous results, data from this study revealed the potential use of WBV in clinical rehabilitation in children with CP. Future investigations should focus on finding the combination of treatment frequency and duration to achieve an ideal result.

PMID: 25461434 [PubMed - as supplied by publisher]

19. Phys Med Rehabil Clin N Am. 2015 Feb;26(1):69-78. doi: 10.1016/j.pmr.2014.09.008.

Pediatric Tone Management.

Vadivelu S1, Stratton A2, Pierce W3.

Tone management is one of the primary roles of a pediatric physiatrist. Hypertonicity frequently inhibits normal movement patterns in children with central nervous system lesions but at times can reinforce muscle group firing and be useful for a child's function. Treatment approaches should be individualized based on functional goals, degree of impairment, interference with care, and type and location of hypertonicity. Treatment plans should be created in collaboration with all individuals caring for the child. There are many causes of hypertonicity as well as many nonsurgical and surgical treatments. Historical and current evidence-based treatments are reviewed.

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20. Res Dev Disabil. 2014 Oct 20;36C:134-141. doi: 10.1016/j.ridd.2014.09.013. [Epub ahead of print]

Humeral external rotation handling by using the Bobath concept approach affects trunk extensor muscles electromyography in children with cerebral palsy.

Grazziotin Dos Santos C1, Pagnussat AS2, Simon AS3, Py R3, Pinho AS4, Wagner MB5.

This study aimed to investigate the electromyographic activity of cervical and trunk extensors muscles in children with cerebral palsy during two handlings according to the Bobath concept. A crossover trial involving 40 spastic

diplegic children was conducted. Electromyography (EMG) was used to measure muscular activity at sitting position (SP), during shoulder internal rotation (IR) and shoulder external rotation (ER) handlings, which were performed using the elbow joint as key point of control. Muscle recordings were performed at the fourth cervical (C4) and at the tenth thoracic (T10) vertebral levels. The Gross Motor Function Classification System (GMFCS) was used to assess whether muscle activity would vary according to different levels of severity. Humeral ER handling induced an increase on EMG signal of trunk extensor muscles at the C4 (P=0.007) and T10 (P<0.001) vertebral levels. No significant effects were observed between SP and humeral IR handling at C4 level; However at T10 region, humeral IR handling induced an increase of EMG signal (P=0.019). Humeral ER resulted in an increase of EMG signal at both levels, suggesting increase of extensor muscle activation. Furthermore, the humeral ER handling caused different responses on EMG signal at T10 vertebra level, according to the GMFCS classification (P=0.017). In summary, an increase of EMG signal was observed during ER handling in both evaluated levels, suggesting an increase of muscle activation. These results indicate that humeral ER handling can be used for diplegic CP children rehabilitation to facilitate cervical and trunk extensor muscles activity in a GMFCS level-dependent manner.

PMID: 25462474 [PubMed - as supplied by publisher]

21. Phys Med Rehabil Clin N Am. 2015 Feb;26(1):89-93. doi: 10.1016/j.pmr.2014.09.011.

Intrathecal Baclofen Bolus Dosing and Catheter Tip Placement in Pediatric Tone Management.

Skalsky AJ1, Fournier CM2.

Intrathecal baclofen (ITB), administered by an implanted pump, has emerged as an efficacious therapy for the treatment of hypertonicity in pediatrics. Although ITB has been used for more than 20 years clinically, much is still unknown about the most optimal dosing regimens and intrathecal catheter tip placement. Clinician experience, animal research, and advanced imaging is guiding the use of ITB. The rationale for high cervical catheter tip placement and pulsating flex dosing is described.

PMID: 25479782 [PubMed - as supplied by publisher]

22. Arch Phys Med Rehabil. 2014 Nov 6. pii: S0003-9993(14)01214-3. doi: 10.1016/j.apmr.2014.10.013. [Epub ahead of print]

Non-invasive Brain Stimulation: The Potential for Use in the Rehabilitation of Pediatric Acquired Brain Injury.

Chung M1, Lo W2.

Non-invasive brain stimulation (NIBS) offers the potential to modulate neural activity and recovery after acquired brain injury. There are few studies of NIBS in children, but a survey of those studies might provide insights into the potential for NIBS to modulate motor rehabilitation, seizure, and behavior in children. We surveyed the published literature prior to July 2014 for articles pertaining to children and NIBS with a focus on case series or trials. We also reviewed selected articles involving adults to illustrate specific points where the literature in children is lacking. A limited number of articles suggest that NIBS can transiently improve motor function. The evidence for an effect on seizures is mixed. Two open-label studies reported improvement of mood in adolescents with depression. NIBS may serve as a tool for pediatric neuro-rehabilitation, but many gaps in our knowledge must be filled before NIBS can be adopted as a clinical intervention. To move forward, the field needs adequately powered trials that can answer these questions. Such trials will be challenging to perform, will likely require multi-center collaboration, and may need to adopt novel trial designs that have been used with rare disorders.

PMID: 25448248 [PubMed - as supplied by publisher]

23. Interaction of BCI with the underlying neurological conditions in patients: pros and cons.

Vuckovic A1, Pineda JA2, LaMarca K3, Gupta D4, Guger C5.

PMID: 25477814 [PubMed - as supplied by publisher]

24. Neurobiol Dis. 2014 Oct 28. pii: S0969-9961(14)00327-1. doi: 10.1016/j.nbd.2014.10.014. [Epub ahead of print]

Brain-controlled muscle stimulation for the restoration of motor function.

Ethier C1, Miller LE2.

Loss of the ability to move, as a consequence of spinal cord injury or neuromuscular disorder, has devastating consequences for the paralyzed individual, and great economic consequences for society. Functional electrical stimulation (FES) offers one means to restore some mobility to these individuals, improving not only their autonomy, but potentially their general health and well-being as well. FES uses electrical stimulation to cause the paralyzed muscles to contract. Existing clinical systems require the stimulation to be preprogrammed, with the patient typically using residual voluntary movement of another body part to trigger and control the patterned stimulation. The rapid development of neural interfacing in the past decade offers the promise of dramatically improved control for these patients, potentially allowing continuous control of FES through signals recorded from motor cortex, as the patient attempts to control the paralyzed body part. While application of these 'brain-machine interfaces' (BMIs) has undergone dramatic development for control of computer cursors and even robotic limbs, their use as an interface for FES has been much more limited. In this review, we consider both FES and BMI technologies and discuss the prospect for combining the two to provide important new options for paralyzed individuals.

PMID: 25447224 [PubMed - as supplied by publisher]

25. Ann Phys Rehabil Med. 2014 Oct 18. pii: S1877-0657(14)01798-9. doi: 10.1016/j.rehab.2014.09.012. [Epub ahead of print]

Quality of life and satisfaction after multilevel surgery in cerebral palsy: Confronting the experience of children and their parents.

Stephan-Carlier A1, Facione J2, Speyer E3, Rumilly E4, Paysant J5.

OBJECTIVES: If the benefits of single-event multilevel surgery (SEMS) in ambulatory children with cerebral palsy have already been validated, especially in terms of functional outcomes, fewer studies have evaluated SEMS in terms of quality of life and satisfaction with surgical outcomes, especially pertaining to the opinions of children and their family. The objectives of this study were to confront the perceptions of parents and the experience of their operated children in terms of quality of life and surgical outcomes. MATERIALS AND Methods:This was an observational, descriptive, single-center study conducted in a regional Pediatric Physical Medicine and Rehabilitation (PM&R) center, which is considered a reference center in this region of France. The subjects recruited were ambulatory children with cerebral palsy (CP) who had SEMS between 2009 and 2011, and one of their parents. The specific "child" and "parent" CP-related modules of the DISABKIDS questionnaire were used to assess these children's quality of life post-surgery. Perceptions of parents and children regarding gait evolution and satisfaction with surgical outcomes were analyzed via a questionnaire developed by the author. Regarding quality of life and surgical outcomes, the correlation between the perception of parents and experience of children was estimated using the intraclass correlation coefficient (ICC) or prevalence-adjusted bias-adjusted kappa values (PABAK). RESULTS: Twelve children (83% of them boys) and their parents participated in the study. For the DISABKIDS questionnaire, a moderate correlation was found regarding functional impact (ICC=0.58; P<0.0178) but a high correlation was reported for the communication item of the questionnaire (ICC=0.73; P=0.0025). Regarding satisfaction with surgical outcomes, results showed a good correlation (PABAK=0.64). Conclusion: Concerning quality of life and satisfaction with surgical outcomes, our study showed a good or even high correlation between parents' perceptions and their child's experience. It is however essential to privilege the child's opinion whenever

possible.

PMID: 25455025 [PubMed - as supplied by publisher]

26. PM R. 2014 Nov 20. pii: S1934-1482(14)01491-9. doi: 10.1016/j.pmrj.2014.11.002. [Epub ahead of print]

Comparative effects of multi-level muscle tendon surgery, osteotomies and dorsal rhizotomy on functional and gait outcome measures for children with cerebral palsy.

Feger MA1, Lunsford CD2, Sauer LD2, Abel MF2, Novicoff W2.

OBJECTIVE: To compare the impact of common surgical interventions including selective dorsal rhizotomy, muscle -tendon surgery and osteotomies for patients with cerebral palsy on Gross Motor Function Measure and temporal, kinematic and kinetic gait variables as assessed via 3D motion analysis. DESIGN: Retrospective cohort study SETTING: Motion analyses laboratory PARTICIPANTS: Ninety-four patients with cerebral palsy, 56 undergoing surgery (37 - MTS, 11 - osteotomy, 8 - SDR) and 38 followed without surgery, ages four to eighteen years old with a GMFCS classification of I, II or III. INTERVENTIONS: Single-event, multi-level muscle tendon surgery, selective dorsal rhizotomy and osteotomy MAIN OUTCOME MEASURES: Change scores (Post intervention - Pre intervention) in Gross Motor Function measure and temporal, kinematic and kinetic gait variables RESULTS: No statistically significant differences in change scores were found between groups in the Gross Motor Function Measure, velocity or stride length measures following the observation period. The selective dorsal rhizotomy group had greater improvements in knee extension when compared to the non-surgical group and greater hip and knee total range of motion during the gait cycle when compared with controls, muscle-tendon surgery and osteotomy cohorts. Lastly, the muscle-tendon surgery group had greater improvements in total knee range of motion compared to the non-surgical group. Conclusion: We have presented evidence that patients who undergo selective dorsal rhizotomy and, to a lesser extent, muscle tendon procedures demonstrate greater improvements in kinematic gait variables over non-surgical interventions in patients with spasticity from cerebral palsy.

PMID: 25460211 [PubMed - as supplied by publisher]

27. Res Dev Disabil. 2014 Nov 29;37C:127-134. doi: 10.1016/j.ridd.2014.11.012. [Epub ahead of print]

Predictors for the benefit of selective dorsal rhizotomy.

Funk JF1, Panthen A2, Bakir MS3, Gruschke F3, Sarpong A2, Wagner C4, Lebek S3, Haberl EJ5.

Selective dorsal rhizotomy (SDR) is a spasticity reducing treatment option for children with spastic cerebral palsy. Selection criteria for this procedure are inconclusive to date. Clinical relevance of the achieved functional improvements and side effects like the negative impact on muscle strength are discussed controversially. In this prospective cohort study one and two year results of 54 children with a mean age of 6.9 (±2.9) years at the time of SDR are analyzed with regard to gross motor function and factors affecting the functional benefit. Only ambulatory children who were able to perform a gross motor function measure test (GMFM-88) were included in this study. Additionally, the modified Ashworth scale (MAS), a manual muscle strength test (MFT), and the body mass index (BMI) were evaluated as possible outcome predictors. MAS of hip adductors and hamstrings decreased significantly ($p<0.001$) and stayed reduced after two years, while GMFM improved significantly from 79% to 84% 12 months after SDR ($p<0.001$) and another 2% between 12 and 24 months ($p=0.002$). Muscle strength did improve significantly concerning knee extension ($p=0.008$) and ankle dorsiflexion ($p=0.006$). The improvement of function correlated moderately with age at surgery and preoperative GMFM and weakly with the standard deviation score of the BMI, the dorsiflexor and plantarflexor strength preoperatively as well as with the reduction of spasticity of the hamstrings and the preoperative spasticity of the adductors and hamstrings. Correctly indicated SDR reduces spasticity and increases motor skills sustainably in children with spastic cerebral palsy corresponding to clinically relevant changes of GMFM without compromising muscular strength. Outcome correlates to GMFM and age rather than to MAS and maximal strength testing. The data of this evaluation suggest that children who benefit the most

from SDR are between 4 and 7 years old and have a preoperative GMFM between 65% and 85%.

PMID: 25460226 [PubMed - as supplied by publisher]

28. Pediatr Neurol. 2014 Oct 13. pii: S0887-8994(14)00600-6. doi: 10.1016/j.pediatrneurol.2014.10.003. [Epub ahead of print]

"Growing" Cerebellum in an Infant After Shunt Insertion.

Benvenisti H1, Bassan H2, Shiran S3, Constantini S4, Roth J5.

INTRODUCTION: Supratentorial cortical mantle growth after shunt surgery in infants with posthemorrhagic hydrocephalus is common. However, cerebellar growth and Chiari are rare. PATIENT DESCRIPTION: We describe a term newborn with an intraventricular hemorrhage and posthemorrhagic hydrocephalus who underwent endoscopic third ventriculostomy followed by shunt placement at age 4 months. RESULTS: After shunt placement, her head circumference growth rate rapidly decreased from the ninety-seventh percentile to the third percentile. Six months after a shunt placement, cerebellar disproportional growth was noticed. Five years after surgery, her cerebellar volume had increased by 300% whereas the cerebral hemispheres volume by 150%, and Chiari 1 appeared. She manifested early hemiparetic cerebral palsy, but, did not develop clinical evidence of increased intracranial pressure or brainstem abnormalities. Conclusion: This term newborn exhibited apparent cerebellar "growth" and posterior fossa crowding after shunt surgery for posthemorrhagic hydrocephalus. Our patient's findings may have resulted from shunt-related alterations in pressure dynamics, leading to decreased head growth rate with a relatively smaller posterior fossa, in face of a normal brain growth. The timing of intraventricular hemorrhage at term, beyond the vulnerable period of cerebellar development, may have been a contributing factor to the craniocerebellar disproportion and posterior fossa crowding cerebellar development may have been relatively spared and was a contributing factor to the craniocerebellar disproportion and posterior fossa crowding.

PMID: 25468532 [PubMed - as supplied by publisher]

29. J Neurol Neurosurg Psychiatry. 2014 Nov 28. pii: jnnp-2014-309255. doi: 10.1136/jnnp-2014-309255. [Epub ahead of print]

Physiotherapy for functional motor disorders: a consensus recommendation.

Nielsen G1, Stone J2, Matthews A3, Brown M3, Sparkes C4, Farmer R5, Masterton L6, Duncan L6, Winters A2, Daniell L2, Lumsden C6, Carson A7, David AS8, Edwards M9.

BACKGROUND: Patients with functional motor disorder (FMD) including weakness and paralysis are commonly referred to physiotherapists. There is growing evidence that physiotherapy is an effective treatment, but the existing literature has limited explanations of what physiotherapy should consist of and there are insufficient data to produce evidence-based guidelines. We aim to address this issue by presenting recommendations for physiotherapy treatment. Methods: A meeting was held between physiotherapists, neurologists and neuropsychiatrists, all with extensive experience in treating FMD. A set of consensus recommendations were produced based on existing evidence and experience. RESULTS: We recommend that physiotherapy treatment is based on a biopsychosocial aetiological framework. Treatment should address illness beliefs, self-directed attention and abnormal habitual movement patterns through a process of education, movement retraining and self-management strategies within a positive and non-judgemental context. We provide specific examples of these strategies for different symptoms. Conclusion: Physiotherapy has a key role in the multidisciplinary management of patients with FMD. There appear to be specific physiotherapy techniques which are useful in FMD and which are amenable to and require prospective evaluation. The processes involved in referral, treatment and discharge from physiotherapy should be considered carefully as a part of a treatment package.

PMID: 25433033 [PubMed - as supplied by publisher] Free full text

30. BMC Pediatr. 2014 Dec 5;14(1):301. [Epub ahead of print]

The impact of hospital-based and community based models of cerebral palsy rehabilitation: a quasi-experimental study.

Dambi JM, Jelsma J.

Background: Cerebral palsy requires appropriate on-going rehabilitation intervention which should effectively meet the needs of both children and parents/care-givers. The provision of effective support is a challenge, particularly in resource constrained settings. A quasi-experimental pragmatic research design was used to compare the impact of two models of rehabilitation service delivery currently offered in Harare, Zimbabwe, an outreach-based programme and the other institution-based. Method: Questionnaires were distributed to 46 caregivers of children with cerebral palsy at baseline and after three months. Twenty children received rehabilitation services in a community setting and 26 received services as outpatients at a central hospital. The Gross Motor Function Measurement was used to assess functional change. The burden of care was measured using the Caregiver Strain Index, satisfaction with physiotherapy was assessed using the modified Medrisk satisfaction with physiotherapy services questionnaire and compliance was measured as the proportion met of the scheduled appointments. Results: Children receiving outreach-based treatment were significantly older than children in the institution-based group. Regression analysis revealed that, once age and level of severity were controlled for, children in the outreach-based treatment group improved their motor function 6% more than children receiving institution-based services.There were no differences detected between the groups with regard to caregiver well-being and 51% of the caregivers reported signs consistent with clinical distress/depression. Most caregivers (83%) expressed that they were overwhelmed by the caregiving role and this increased with the chronicity of care. The financial burden of caregiver was predictive of caregiver strain.Caregivers in the outreach-based group reported greater satisfaction with services and were more compliant (p¿<¿.001) as compared to recipients of institution-based services. Conclusion: Long term caregiving leads to strain in caregivers and there is a need to design interventions to alleviate the burden. The study was a pragmatic, quasi-experimental study thus causality cannot be inferred. However findings from this study suggest that the provision of care within a community setting as part of a well-structured outreach programme may be preferable method of service delivery within a resource-constrained context. It was associated with a greater improvement in functioning, greater satisfaction with services and better compliance.

PMID: 25476072 [PubMed - as supplied by publisher]

31. Pediatrics. 2014 Dec;134(6):1222. doi: 10.1542/peds.2014-3029.

Whittingham et al. Interventions to Reduce Behavioral Problems in Children With Cerebral Palsy: An RCT. Pediatrics. 2014;133(5):e1249-e1257.

[No authors listed]

PMID: 25452641 [PubMed - in process]

32. Res Dev Disabil. 2014 Nov 27;37C:102-111. doi: 10.1016/j.ridd.2014.11.005. [Epub ahead of print]

Predictors of participation change in various areas for preschool children with cerebral palsy: A longitudinal study.

Wu KP1, Chuang YF2, Chen CL3, Liu IS4, Liu HT5, Chen HC6.

This study identifies potential predictors of participation changes in various areas for preschool children with cerebral palsy (CP). Eighty children with CP (2-6 years) were enrolled. Seven potential predictors were identified: age; sex; socioeconomic status, CP subtype; cognitive function, Function Independence Measure for Children (WeeFIM), and motor composite variable from 5 motor factors (gross motor function classification system (GMFCS) level; bimanual fine motor function level; selective motor control score; Modified Ashworth Scale score; and Spinal

Alignment and Range of Motion Measure). Outcome was assessed at baseline and at 6-month follow-up using the Assessment of Preschool Children's Participation (APCP) including diversity and intensity scores in the areas of play (PA), skill development (SD), active physical recreation, social activities (SA), and total areas. Dependent variables were change scores of APCP scores at baseline and 6-month follow-up. Regression analyses shows age and sex together predicted for APCP-total, APCP-SD diversity and APCP-total intensity changes ($r2=0.13-0.25$, $p<0.001$); cognitive function and WeeFIM were negative predictors for APCP-SA and APCP-PA diversity changes, respectively. CP subtype, motor composite variable, and socioeconomic status predicted for APCP changes in some areas. Findings suggest that young boys with poor cognitive function and daily activity predicted most on participation changes.

PMID: 25460224 [PubMed - as supplied by publisher]

33. Res Dev Disabil. 2014 Nov 14;36C:551-564. doi: 10.1016/j.ridd.2014.10.043. [Epub ahead of print]

Predictors of participation of adolescents with cerebral palsy: A European multi-centre longitudinal study.

Dang VM1, Colver A2, Dickinson HO3, Marcelli M4, Michelsen SI5, Parkes J6, Parkinson K7, Rapp M8, Arnaud C9, Nystrand M10, Fauconnier J11.

We investigated whether childhood factors that are amenable to intervention (parenting stress, child psychological problems and pain) predicted participation in daily activities and social roles of adolescents with cerebral palsy (CP). We randomly selected 1174 children aged 8-12 years from eight population-based registers of children with CP in six European countries; 743 (63%) agreed to participate. One further region recruited 75 children from multiple sources. These 818 children were visited at home at age 8-12 years, 594 (73%) agreed to follow-up at age 13-17 years. We used the following measures: parent reported stress (Parenting Stress Index Short Form), their child's psychological difficulties (Strength and Difficulties Questionnaire) and frequency and severity of pain; either child or parent reported the child's participation (LIFE Habits questionnaire). We fitted a structural equation model to each of the participation domains, regressing participation in childhood and adolescence on parenting stress, child psychological problems and pain, and regressing adolescent factors on the corresponding childhood factors; models were adjusted for impairment, region, age and gender. Pain in childhood predicted restricted adolescent participation in all domains except Mealtimes and Communication (standardized total indirect effects ß -0.05 to -0.18, $0.01<p<0.05$ to $p<0.001$, depending on domain). Psychological problems in childhood predicted restricted adolescent participation in all domains of social roles, and in Personal Care and Communication (ß -0.07 to -0.17, $0.001<p<0.01$ to $p<0.001$). Parenting stress in childhood predicted restricted adolescent participation in Health Hygiene, Mobility and Relationships (ß -0.07 to -0.18, $0.001<p<0.01$ to $p<0.001$). These childhood factors predicted adolescent participation largely via their effects on childhood participation; though in some domains early psychological problems and parenting stress in childhood predicted adolescent participation largely through their persistence into adolescence. We conclude that participation of adolescents with CP was predicted by early modifiable factors related to the child and family. Interventions for reduction of pain, psychological difficulties and parenting stress in childhood are justified not only for their intrinsic value, but also for probable benefits to childhood and adolescent participation.

PMID: 25462516 [PubMed - as supplied by publisher] Free full text

34. J Crit Care. 2014 Oct 25. pii: S0883-9441(14)00423-7. doi: 10.1016/j.jcrc.2014.10.016. [Epub ahead of print]

Adults with childhood-onset chronic conditions admitted to US pediatric and adult intensive care units.

Edwards JD1, Vasilevskis EE2, Yoo EJ3, Houtrow AJ4, Boscardin WJ5, Dudley RA6, Okumura MJ7.

PURPOSE: The purpose of the study is to compare demographics, intensive care unit (ICU) admission characteristics, and ICU outcomes among adults with childhood-onset chronic conditions (COCCs) admitted to US pediatric and adult ICUs. MATERIALS AND Methods: Retrospective cross-sectional analyses of 6088 adults aged

19 to 40 years admitted in 2008 to 70 pediatric ICUs that participated in the Virtual Pediatric Intensive Care Unit Performance Systems and 50 adult ICUs that participated in Project IMPACT. RESULTS: Childhood-onset chronic conditions were present in 53% of young adults admitted to pediatric units, compared with 9% of those in adult units. The most common COCC in both groups were congenital cardiac abnormalities, cerebral palsy, and chromosomal abnormalities. Adults with COCC admitted to pediatric units were significantly more likely to be younger, have lower functional status, and be nontrauma patients than those in adult units. The median ICU length of stay was 2 days, and the intensive care unit mortality rate was 5% for all COCC patients with no statistical difference between pediatric or adult units. Conclusion: There are marked differences in characteristics between young adults with COCC admitted to pediatric ICUs and adult ICUs. Barriers to accommodating these young adults may be reasons why many such adults have not transitioned from pediatric to adult critical care.

PMID: 25466316 [PubMed - as supplied by publisher]

35. Phys Med Rehabil Clin N Am. 2015 Feb;26(1):39-56. doi: 10.1016/j.pmr.2014.09.001.

The Importance of Good Nutrition in Children with Cerebral Palsy.

Rempel G.

Poor nutritional status, which is common in children with cerebral palsy (CP), has generated considerable interest because of its wide-ranging impact on the children's health and well-being. Understanding the causes of poor nutrition, and the appropriate measurements required to interpret the nutritional status in children with CP, are integral to developing appropriate nutritional intervention strategies. Focusing attention on improving nutrition early in the lives of children with CP affords families and care providers with a unique opportunity for intervention, which may result in better outcomes for the children.

PMID: 25479778 [PubMed - as supplied by publisher]

36. Int J Surg Case Rep. 2014 Nov 4;5(12):1072-1073. doi: 10.1016/j.ijscr.2014.10.072. [Epub ahead of print]

An unusual complication of Botox treatment for sialorrhoea.

Layton TB.

INTRODUCTION: To illustrate the potential side effects and clinical efficacy of Botox injections in treating sialorrhoea. PRESENTATION OF CASE: A 26-year-old patient with cerebral palsy with dystonia had a long history of severe, distressing sialorrhoea. She was treated with three separate Botox injections into her salivary glands in December 2011, July 2012 and March 2013. DISCUSSION: Following the Botox injections the patient developed dysphagia, began to expectorate thick mucus and developed a cough; she was treated for a chest injection and during this time her feeding deteriorated. Three injections were given as the patient had an objective and significant reduction in salivation. However, the side effect profile was deemed too great to continue with treatment. Conclusion: Botox is a novel and effective treatment for reducing saliva production. Its clinical efficacy is supported by this case and correlates with the recent literature. Although rare, significant side effects can happen and the case presented illustrates the care needed when administering injections, particularly in a subgroup of patients.

PMID: 25460477 [PubMed - as supplied by publisher] Free full text

37. Br J Ophthalmol. 2014 Dec 3. pii: bjophthalmol-2014-305758. doi: 10.1136/bjophthalmol-2014-305758. [Epub ahead of print]

Ocular alignment after bilateral lateral rectus recession in exotropic children with cerebral palsy.

Han SY1, Han J2, Han SH1, Lee JB2, Rhiu S3.

AIMS: To compare the surgical outcomes of exotropia patients with cerebral palsy (CP) and controls without CP. Methods: 30 patients with exotropia and CP and 60 age-matched controls without CP who underwent bilateral lateral rectus (BLR) recession were retrospectively enrolled. All patients underwent BLR recession according to Parks' method. Surgical success was defined by esotropia deviation =5 PD (prism diopters) and exotropia deviation =10 PD. Success rate, cumulative probabilities of success, and postoperative change of angle deviations using a linear mixed model were evaluated. RESULTS: Mean postoperative follow-up times were 21.60±8.62 months for the CP group and 25.60±10.82 months for the control group (p=0.081). Mean preoperative deviation was 30.97±10.54 PD in the CP group and 29.75±7.52 PD in the control group (p=0.530), which was reduced to 4.44±7.13 PD and 7.43±7.59 PD, respectively (p=0.093) after BLR recession. Using linear mixed model analysis, the estimated mean postoperative deviation of both groups was in the success range at all times tested. At the final visit, successful surgical outcomes were achieved in 76.7% of the CP group and 56.7% of the control group (p=0.179). Differences in the cumulative probability of surgical success were not statistically significant between the two groups (p=0.106, log rank test). Conclusion: CP patients with exotropia showed successful ocular alignment comparable to that of controls after BLR recession of at least 2 years. Published by the BMJ Publishing Group Limited. For permission to use (where not already granted under a licence) please go to http://group.bmj.com/group/rights-licensing/permissions.

PMID: 25472950 [PubMed - as supplied by publisher]

38. Res Dev Disabil. 2014 Nov 20;36C:630-644. doi: 10.1016/j.ridd.2014.10.025. [Epub ahead of print]

Spontaneous improvement in oculomotor function of children with cerebral palsy.

Ego C1, Orban de Xivry JJ2, Nassogne MC3, Yüksel D4, Lefèvre P5.

Eye movements are essential to get a clear vision of moving objects. In the present study, we assessed quantitatively the oculomotor deficits of children with cerebral palsy (CP). We recorded eye movements of 51 children with cerebral palsy (aged 5-16 years) with relatively mild motor impairment and compared their performance with age-matched control and premature children. Overall eye movements of children with CP are unexpectedly close to those of controls even though some oculomotor parameters are biased by the side of hemiplegia. Importantly, the difference in performance between children with CP and controls decreases with age, demonstrating that the oculomotor function of children with CP develops as fast as or even faster than controls for some visual tracking parameters. That is, oculomotor function spontaneously improves over the course of childhood. This evolution highlights the ability of lesioned brain of children with CP to compensate for impaired motor function beyond what would be achieved by normal development on its own.

PMID: 25462523 [PubMed - as supplied by publisher]

39. J Phys Ther Sci. 2014 Nov;26(11):1679-1684. Epub 2014 Nov 13.

Relationship among Ocular Diseases, Developmental Levels, and Clinical Characteristics of Children with Diplegic Cerebral Palsy.

Boyaci A1, Akal A2, Tutoglu A1, Kandemir H3, Koca I4, Boyraz I5, Celen E1, Ozkan U2.

Purpose: The aim of this study was to evaluate the relationships among vision problems, developmental levels, upper extremity functions, and qualities of life of children with cerebral palsy (CP). Subjects: The study included 32 children, aged 4-15 years, diagnosed with diplegic type CP. Methods: Hand function was evaluated using the Manual Ability Classification System (MACS) and the Bimanual Fine Motor Function (BFMF) scale, and the severity

of CP was assessed using the Gross Motor Function Classification System (GMFCS). The developmental and mental capabilities of the children were evaluated using the Ankara Developmental Screening Inventory (ADSI) or the WISC-R test. An oculomotor examination was conducted for all patients. [Results] Positive correlations were found between GMFCS and BFMF, GMFCS and MACS, and MACS and BFMF scores (r=0.636; r=0.553; r=0.718, respectively). Significant correlations were found between upper extremity function, the severity of CP, the quality of life, and the general developmental level. There was no significant correlation between ocular disorders and clinical characteristics. [Conclusion:] GMFCS, MACS, and BFMF may be useful for defining the functional status of children with CP, as they are easy, practical, and simple classification scales that conform to each other.

PMID: 25435676 [PubMed - as supplied by publisher] PMCID: PMC4242931 Free PMC Article

40. Braz J Otorhinolaryngol. 2014 Oct 19. pii: S1808-8694(14)00126-8. doi: 10.1016/j.bjorl.2014.10.002. [Epub ahead of print]

Hearing rehabilitation in cerebral palsy: development of language and hearing after cochlear implantation.

Hilgenberg AM1, Cardoso CC1, Caldas FF1, Tschiedel RD2, Deperon TM3, Bahmad Jr F4.

INTRODUCTION: Auditory rehabilitation in children with bilateral severe-to-profound sensorineural hearing loss with cochlear implant has been developed in recent decades; however, the rehabilitation of children with cerebral palsy still remains a challenge to otolaryngology and speech therapy professionals. OBJECTIVE: To verify the effectiveness of cochlear implants in the development of auditory and language skills in children with cerebral palsy. Methods: A prospective analytical study. The evaluation of auditory responses to speech test was applied to the children in this study at regular intervals following implantation. Standardized tests that assess and quantify the development of auditory and language skills were administered and speech therapy video records and speech therapy files were analyzed. All children went through individually tailored intensive audiological rehabilitation programs following cochlear implantation. RESULTS: Two participants had gradual auditory and language development when compared to other participants who reached advanced levels in hearing and oral language classifications. Conclusion: The use of the Cochlear implant enabled participants to reach advanced stages of hearing and language skills in three of the five participants with cerebral palsy in this study. This electronic device is a viable therapeutic option for children with cerebral palsy to help them achieve complex levels of auditory and language skills.

PMID: 25458256 [PubMed - as supplied by publisher] Free full text

41. J Clin Diagn Res. 2014 Oct;8(10):ND07-ND09. Epub 2014 Oct 20.

Testicular Torsion in Cerebral Palsy - Resident's Grey Area.

Ghalige HS1, N B1, Maibam C2, Sharma MB3, Singh TS3.

The association of testicular torsion and cerebral palsy is a well-known fact. But the infrequent presentation to emergency room makes the clinician sceptical. Such a presentation often puzzles the residents regarding the diagnosis and the treatment. Here we present a case of an adolescent boy aged with cerebral palsy 13 years with incessant crying and not feeding well for last 3days. Right inguinal region showed a tender globular swelling with absence of testis in scrotum along with signs of septicaemia. The inguinal exploration was performed under general anaesthesia which revealed gangrenous right testis. Right orchidectomy and left orchidopexy was performed and the patient recovered well. This case is reported for its complexity due to lack of reliable history, delayed presentation and associated comorbidities posing challenges to the treating surgeons.

PMID: 25478396 [PubMed - as supplied by publisher]

Prevention and Cure

42. Acta Inform Med. 2012 Jan;18(2):64-82. Epub 2012 Jan 1.

Intrauterine Growth Restriction and Cerebral Palsy.

Kurjak A1, Predojevic M2, Stanojevic M3, Kadic AS4, Miskovic B5, Badreldeen A6, Talic A7, Zaputovic S5, Honemeyer U8.

Intrauterine growth restriction (IUGR) can be described as condition in which fetus fails to reach his potential growth. It is common diagnosis in obstetrics, and carries an increased risk of perinatal mortality and morbidity. Moreover, IUGR has lifelong implications on health, especially on neurological outcome. There is a need for additional neurological assessment during monitoring of fetal well-being, in order to better predict antenatally which fetuses are at risk for adverse neurological outcome. Studies have revealed that the behavior of the fetus reflects the maturational processes of the central nervous system (CNS). Hence, ultrasound investigation of the fetal behavior can give us insight into the integrity and functioning of the fetal CNS. Furthermore, investigations carried out using modern method, four-dimensional (4D) sonography, have produced invaluable details of fetal behavior and its development, opening the door to a better understanding of the prenatal functional development of the CNS. Based on previous observations and several years of investigation, our reaserch group has proposed a new scoring system for the assessment of fetal neurological status by 4D sonography named Kurjak antenatal neurodevelopmental test (KANET). The value of KANET in distinguishing fetal brain and neurodevelopmental alterations due to the early brain impairment in utero is yet to be assessed in large population studies. However, preliminary results are very encouraging.

PMID: 25473145 [PubMed - as supplied by publisher]

43. Am J Obstet Gynecol. 2014 Oct 30. pii: S0002-9378(14)02195-4. doi: 10.1016/j.ajog.2014.10.1103. [Epub ahead of print]

Fetal growth restriction and risk of cerebral palsy in singletons born after at least 35 weeks' gestation.

Blair EM1, Nelson KB2.

OBJECTIVE: The objective of the study was to improve the understanding of etiological paths to cerebral palsy (CP) that include fetal growth restriction by examining factors associated with growth restriction that modify CP risk. STUDY DESIGN: In a total population of singletons born at or after 35 weeks, there were 493 children with CP and 508 matched controls for whom appropriateness of fetal growth could be estimated. Fetal growth was considered markedly restricted if birthweight was more than 2 SD below optimal for gender, gestation, maternal height, and parity. We examined maternal blood pressure in pregnancy, smoking, birth asphyxia, and major birth defects recognized by age 6 years as potential modifiers of CP risk in growth-restricted births. RESULTS: More than 80% of term and late preterm markedly growth-restricted singletons were born following a normotensive pregnancy and were at statistically significantly increased risk of CP (odds ratio, 4.81; 95% confidence interval, 2.7-8.5), whereas growth-restricted births following a hypertensive pregnancy were not. Neither a clinical diagnosis of birth asphyxia nor potentially asphyxiating birth events occurred more frequently among growth-restricted than among appropriately grown infants with CP. Major birth defects, particularly cerebral defects, occurred in an increasing proportion of CP with increasing growth deficit. The factor most predictive of CP in growth-restricted singletons was a major birth defect, present in 53% of markedly growth-restricted neonates with later CP. Defects observed in CP were similar whether growth restricted or not, except for an excess of isolated congenital microcephaly in those born growth restricted. The highest observed CP risk was in infants with both growth restriction and a major birth defect (8.9% of total CP in this gestational age group, 0.4% of controls: odds ratio, 30.9; 95% confidence interval, 7.0-136). Conclusion: The risk of CP was increased in antenatally growth-restricted singletons born at or near term to normotensive mothers. In growth-restricted singletons, a major birth defect was the dominant predictor, associated with a 30-fold increase in odds of CP. Identification of birth defects in the growth-restricted fetus or neonate may provide significant prognostic information.

PMID: 25448521 [PubMed - as supplied by publisher]

44. CNS Neurosci Ther. 2014 Dec 5. doi: 10.1111/cns.12365. [Epub ahead of print]

Anti-tissue Plasminogen Activator (tPA) as an Effective Therapy of Neonatal Hypoxia-Ischemia with and without Inflammation.

Yang D1, Kuan CY.

Hypoxic-ischemic brain injury is an important cause of neurodevelopmental deficits in neonates. Intrauterine infection and the ensuing fetal inflammatory responses augment hypoxic-ischemic brain injury and attenuate the efficacy of therapeutic hypothermia. Here, we review evidences from preclinical studies suggesting that the induction of brain parenchymal tissue-type plasminogen activator (tPA) plays an important pathogenic role in these conditions. Moreover, administration of a stable-mutant form of plasminogen activator inhibitor-1 called CPAI confers potent protection against hypoxic-ischemic injury with and without inflammation via different mechanisms. Besides intracerebroventricular injection, CPAI can also be administered into the brain using a noninvasive intranasal delivery strategy, adding to its applicability in clinical use. In sum, the therapeutic potential of CPAI in neonatal care merits further investigation with large-animal models of hypoxia-ischemia and cerebral palsy.

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45. Early Hum Dev. 2014 Oct 21;90(12):851-856. doi: 10.1016/j.earlhumdev.2014.09.007. [Epub ahead of print]

Neurological examination combined with brain MRI or cranial US improves prediction of neurological outcome in preterm infants.

Setänen S1, Lahti K2, Lehtonen L3, Parkkola R4, Maunu J5, Saarinen K6, Haataja L7.

BACKGROUND: The predictive value of the combination of neurological examination and brain magnetic resonance imaging (MRI) or cranial ultrasound (cUS) in preterm infants is not known. AIMS: To study the prognostic value of the combination of neurological examination and brain MRI at term equivalent age (TEA) or serial neonatal cUS in very preterm infants for neurosensory outcome at 2years of corrected age. STUDY DESIGN: A prospective follow-up study. SUBJECTS: A total of 216 very preterm infants (birth weight 1132g [SD 331g]) born in Turku University Hospital, from 2001 to 2006, were included. OUTCOME MEASURES: The Dubowitz neurologic examination and brain MRI were done at TEA, and serial cUS examinations were performed until TEA. The Hammersmith Infant Neurological Examination (HINE) and neurosensory impairments (NSI) were assessed at 2years of corrected age. RESULTS: Of all infants, 163 (76%) had one or more deviant neurological items at TEA, and 32 (15%) had the HINE total score below the 10th percentile at 2years of corrected age. A total of 17 (8%) infants had NSI. Neurological examination at TEA improved the negative and positive predictive values of brain MRI for NSI from 99% to 100%, and from 28% to 35%, respectively, and the negative and positive predictive values of cUS from 97% to 100%, and from 61% to 79%, respectively. Conclusion: The combination of the Dubowitz neurologic examination and the brain MRI at TEA or serial neonatal cUS provides a valuable clinical tool for predicting long-term neurosensory outcome in preterm infants.

PMID: 25463831 [PubMed - as supplied by publisher]

46. Early Hum Dev. 2014 Oct 17;90(12):837-842. doi: 10.1016/j.earlhumdev.2014.08.006. [Epub ahead of print]

Long term developmental outcomes of pre-school age children following laser surgery for twin-to-twin transfusion syndrome.

McIntosh J1, Meriki N1, Joshi A2, Biggs V1, Welsh AW3, Challis D1, Lui K4.

BACKGROUND: Laser therapy is now a well recognised treatment for twin-to-twin transfusion syndrome (TTTS). We investigated the early childhood neurodevelopmental outcome of children post laser treatment for TTTS in our centre. Methods: Children of women who had laser therapy for TTTS between March 2006 and June 2008 were assessed at 30-69months of age with WPPSI-III and a general health questionnaire. Major neurodevelopmental impairment (NDI) was reported as IQ<70 or cerebral palsy (CP). Borderline cognitive impairment was defined by IQ 70-79. RESULTS: Amongst the 37 pregnancies treated, 62 infants were discharged home and the overall foetal survival rate was 84%. A total of 50 children (84%) from 31 pregnancies were assessed. Average age at assessment was 47months. Two children with late treatment of congenital hypothyroidism were excluded. The majority of pregnancies were Quintero Stage III (74%). There was a significant trend for worse outcome with higher Quintero stage. The average gestational age at birth was 32weeks. The majority (39, 78%) of children were found to be neurodevelopmentally normal; 9 (18%) had borderline cognitive development; and 2 (4%) had a major NDI, including one with cerebral palsy (2%). Conclusion: There was a modest level of neurocognitive impairment post laser therapy for TTTS, mainly borderline cognitive development, lesser so major NDI. There was a low incidence of cerebral palsy. Routine developmental and neurological follow-up of these children is recommended.

PMID: 25463829 [PubMed - as supplied by publisher]

47. J Neuroimmunol. 2014 Nov 20. pii: S0165-5728(14)00961-8. doi: 10.1016/j.jneuroim.2014.11.015. [Epub ahead of print]

Targets for adjunctive therapy in pneumococcal meningitis.

Barichello T1, Collodel A2, Generoso JS2, Simões LR2, Moreira AP2, Ceretta RA2, Petronilho F3, Quevedo J4.

Pneumococcal meningitis is a severe infectious disease of the central nervous system (CNS) and a significant cause of morbidity and mortality worldwide. The inflammatory reaction to the disease contributes to neuronal injury and involves the meninges, the subarachnoid space and the brain parenchymal vessels. Bacterial pathogens may reach the blood-brain barrier and be recognized by antigen-presenting cells through the binding of Toll-like receptors, triggering an inflammatory cascade. This in turn produces cytokines and chemokines, increases adhesion molecule expression and attracts leukocytes from the blood. This cascade leads to lipid peroxidation, mitochondrial damage and blood-brain barrier permeability. In spite of effective antibacterial treatments, approximately one third of survivors suffer from long-term sequelae, such as hearing loss, cerebral palsy, seizures, hydrocephaly or cognitive impairment. This review summarizes the information on targets of adjuvant treatments of acute pneumococcal meningitis.

PMID: 25468775 [PubMed - as supplied by publisher]

48. J Pediatr (Rio J). 2014 Oct 16. pii: S0021-7557(14)00143-0. doi: 10.1016/j.jped.2014.08.004. [Epub ahead of print]

Systematic review: hereditary thrombophilia associated to pediatric strokes and cerebral palsy.

Torres VM1, Saddi VA2.

OBJECTIVES: This review aimed to organize and consolidate the latest knowledge about mutations and genetic polymorphisms related to hereditary thrombophilia and their potential association with pediatric stroke and cerebral

palsy (CP). SOURCES: Scientific articles published from 1993 to 2013, written in Portuguese, English, French, and Spanish, were selected and reviewed. The publications were searched in electronic databases, and also in the collections of local libraries. The terms "hereditary thrombophilia", "polymorphisms", "mutation", "pediatric strokes", and "cerebral palsy" were used for the research. SUMMARY OF THE FINDINGS: The search in databases and in the bibliographic references retrieved 75 articles for inclusion in this review. Studies that investigated hereditary thrombophilias and their associations to CP and arterial and venous pediatric stroke presented contradictory results. The meta-analysis and case-control studies that showed positive results for this association described only slightly increased relative risks and sometimes had questionable Conclusion: The association of two or more hereditary thrombophilias, or the association between thrombophilia and other specific clinical risk factors, suggest a higher risk of CP and pediatric stroke than isolated hereditary thrombophilia. Conclusion: Larger, multicenter studies should be developed in order to elucidate the role of mutations leading to hereditary thrombophilia and the development of CP and pediatric stroke. The complex and multifactorial etiology of CP and stroke makes this an arduous and difficult task; however, the benefits generated by these studies are immeasurable.

PMID: 25451211 [PubMed - as supplied by publisher] Free full text

49. J Reprod Immunol. 2014 Aug 17;106C:21-26. doi: 10.1016/j.jri.2014.08.001. [Epub ahead of print]

Health and disease in children born after assistive reproductive therapies (ART).

Hyrapetian M1, Loucaides EM2, Sutcliffe AG3.

In vitro fertilisation (IVF) and other assisted reproductive therapies (ART) offer hope to subfertile couples worldwide. At least 5 million ART children have been born to date. Their health is an issue that is increasingly relevant: first, to those children and young adults themselves; second, to couples considering fertility treatment; and third, to the general population as ART has progressed from experimental treatment to routine practice. Many concerns about the potential risks to these children have been voiced with varying degrees of supportive evidence. This article summarises some key long-term data. Current evidence suggests that ART does increase risk of: higher order pregnancy (with its inherent pre- and perinatal risks); prematurity and low birth weight; congenital malformations in particular of the male urogenital system; imprinting disorders. Reassuringly, evidence points away from an increased overall cancer risk or differences in neurodevelopmental outcomes. Many unknowns remain, including future fertility and cardiovascular risks and risk of cerebral palsy.

PMID: 25438931 [PubMed - as supplied by publisher]

50. Orphanet J Rare Dis. 2014 Nov 30;9(1):197. [Epub ahead of print]

Treatable inborn errors of metabolism presenting as cerebral palsy mimics: systematic literature review.

Leach EL, Shevell M, Bowden K, Stockler-Ipsiroglu S, van Karnebeek C.

BackgroundInborn errors of metabolism (IEMs) have been anecdotally reported in the literature as presenting with features of cerebral palsy (CP) or misdiagnosed as `atypical CP¿. A significant proportion is amenable to treatment either directly targeting the underlying pathophysiology (often with improvement of symptoms) or with the potential to halt disease progression and prevent/minimize further damage.Methods: We performed a systematic literature review to identify all reports of IEMs presenting with CP-like symptoms before 5 years of age, and selected those for which evidence for effective treatment exists.ResultsWe identified 54 treatable IEMs reported to mimic CP, belonging to 13 different biochemical categories. A further 13 treatable IEMs were included, which can present with CP-like symptoms according to expert opinion, but for which no reports in the literature were identified. For 26 of these IEMs, a treatment is available that targets the primary underlying pathophysiology (e.g. neurotransmitter supplements), and for the remainder (n¿=¿41) treatment exerts stabilizing/preventative effects (e.g. emergency regimen). The total number of treatments is 50, and evidence varies for the various treatments from Level 1b, c (n¿=¿2); Level 2a, b, c (n¿=¿16); Level 4 (n¿=¿35); to Level 4¿5 (n¿=¿6); Level 5 (n¿=¿8). Thirty-eight (57%) of the treatable IEMs mimicking CP can be identified by ready available metabolic screening tests in blood or urine,

while the remaining IEMs require more specific and sometimes invasive tests. Conclusion: sLimited by the rare nature of IEMs and incomplete information in the literature, we conclude that (1) A surprisingly large number of IEMs can present with CP symptoms, as `CP mimics¿, (2) Although individually rare, a large proportion of these diseases are treatable such that neurological damage can either be reversed or prevented, (3) clinician awareness of treatable CP mimics is important for appropriate screening, diagnosis, and early intervention, and (4) systematic studies are required to elucidate the collective frequency of treatable IEMs in CP.

PMID: 25433678 [PubMed - as supplied by publisher] Free full text

51. Pediatr Neurol. 2014 Oct 16. pii: S0887-8994(14)00603-1. doi: 10.1016/j.pediatrneurol.2014.10.005. [Epub ahead of print]

The Breadth and Type of Systemic Inflammation and the Risk of Adverse Neurological Outcomes in Extremely Low Gestation Newborns.

Kuban KC1, O'Shea MT2, Allred EN3, Fichorova RN4, Heeren T5, Paneth N6, Hirtz D7, Dammann O8, Leviton A9; ELGAN Study Investigators.

BACKGROUND: We hypothesized that the risk of brain damage in extremely preterm neonates increases with the breadth and type of systemic inflammation, indexed by the number of elevated inflammation-related proteins and the number of functional categories of inflammation-related proteins exhibiting an elevated concentration. Methods: In blood from 881 infants born before 28 weeks gestation, we measured the concentrations of 25 inflammation-related proteins, representing six functional categories (cytokines, chemokines, growth factors, adhesion molecules, metalloproteinases, and liver-produced acute phase reactant proteins) on postnatal days 1, 7, and 14. We evaluated associations between the number and type of proteins whose concentrations were elevated on two separate occasions a week apart and the diagnoses of ventriculomegaly as a neonate, and at 2 years, microcephaly, impaired early cognitive functioning, cerebral palsy, and autism risk as assessed with the Modified Checklist for Autism in Toddlers screen, and in a subset of these children from 12 of 14 sites (n = 826), an attention problem identified with the Child Behavior Checklist. RESULTS: The risk of abnormal brain structure and function overall was increased among children who had recurrent and/or persistent elevations of the 25 proteins. The risk for most outcomes did not rise until at least four proteins in at least two functional categories were elevated. When we focused our analysis on 10 proteins previously found to be associated consistently with neurological outcomes, we found the risk of low Mental Development Index on the Bayley Scales of Infant Development-II, microcephaly, and a Child Behavior Checklist-defined attention problem increased with higher numbers of these recurrently and/or persistently elevated proteins. INTERPRETATION: Increasing breadth of early neonatal inflammation, indexed by the number of protein elevations or the number of protein functional classes elevated, is associated with increasing risk of disorders of brain structure and function among infants born extremely preterm.

PMID: 25459361 [PubMed - as supplied by publisher]

52. Pediatr Neurol. 2014 Nov;51(5):657-62. doi: 10.1016/j.pediatrneurol.2014.08.010. Epub 2014 Aug 27.

Erythropoietin and hypothermia for hypoxic-ischemic encephalopathy.

Rogers EE1, Bonifacio SL1, Glass HC2, Juul SE3, Chang T4, Mayock DE3, Durand DJ5, Song D6, Barkovich AJ7, Ballard RA1, Wu YW8.

BACKGROUND: Erythropoietin is neuroprotective in animal models of neonatal hypoxic-ischemic encephalopathy. We previously reported a phase I safety and pharmacokinetic study of erythropoietin in neonates. This article presents the neurodevelopmental follow-up of infants who were enrolled in the phase I clinical trial. Methods: We enrolled 24 newborns with hypoxic-ischemic encephalopathy in a dose-escalation study. Patients received up to six doses of erythropoietin in addition to hypothermia. All infants underwent neonatal brain magnetic resonance imaging (MRI) reviewed by a single neuroradiologist. Moderate-to-severe neurodevelopmental disability was defined as cerebral palsy with Gross Motor Function Classification System levels III-V or cognitive impairment based on Bayley Scales of Infant Development II mental developmental index or Bayley III cognitive composite score.

RESULTS: Outcomes were available for 22 of 24 infants, at mean age 22 months (range, 8-34 months). There were no deaths. Eight (36%) had moderate-to-severe brain injury on neonatal MRI. Moderate-to-severe disability occurred in one child (4.5%), in the setting of moderate-to-severe basal ganglia and/or thalamic injury. Seven infants with moderate-to-severe watershed injury exhibited the following outcomes: normal (three), mild language delay (two), mild hemiplegic cerebral palsy (one), and epilepsy (one). All 11 patients with a normal brain MRI had a normal outcome. Conclusion: This study is the first to describe neurodevelopmental outcomes in infants who received high doses of erythropoietin and hypothermia during the neonatal period. The findings suggest that future studies are warranted to assess the efficacy of this new potential neuroprotective therapy.

PMID: 25439577 [PubMed - in process]

53. Pediatr Neurol. 2014 Dec;51(6):760-768. doi: 10.1016/j.pediatrneurol.2014.07.031. Epub 2014 Aug 14.

Perinatal Arterial Ischemic Stroke: Presentation, Risk Factors, Evaluation, and Outcome.

Lehman LL1, Rivkin MJ2.

BACKGROUND: Perinatal arterial ischemic stroke is as common as large vessel arterial ischemic stroke in adults and leads to significant morbidity. Perinatal arterial ischemic stroke is the most common identifiable cause of cerebral palsy and can lead to cognitive and behavioral difficulties that are amortized over a lifetime. Methods: The literature on perinatal arterial ischemic stroke was reviewed and analyzed. RESULTS: Risk factors for perinatal arterial ischemic stroke include those that are maternal, neonatal, and placental. The most common clinical signs at presentation are seizures and hemiparesis. Evaluation should begin with thorough history acquisition and physical examination followed by magnetic resonance imaging of the brain, with consideration of magnetic resonance angiography of the head and neck, echocardiogram, and thrombophilia evaluation. Treatment beginning early to include physical, speech, and occupational therapies including constraint-induced movement therapy and close cognitive and developmental follow-up may be beneficial. Future treatments may include transcranial magnetic stimulation, hypothermia, and erythropoietin. Conclusion: Perinatal arterial ischemic stroke comprises a group of arterial ischemic injuries that can occur in the prenatal, perinatal, and postnatal periods in term and preterm infants with different types of perinatal arterial ischemic stroke having different clinical presentations, risk factors, and long-term outcomes.

PMID: 25444092 [PubMed - as supplied by publisher]

54. Pol J Radiol. 2014 Nov 30;79:444-449. eCollection 2014.

Diagnostic Imaging and Problems of Schizencephaly.

Stopa J1, Kucharska-Miasik I1, Dziurzynska-Bialek E1, Kostkiewicz A1, Solinska A2, Zajac-Mnich M2, Guz W2, Samojedny A2.

BACKGROUND: Schizencephaly is a rare developmental malformation of the central nervous system associated with cell migration disturbances. Schizencephaly can be uni- or bilateral and is divided into two morphological types. The cleft is defined as type I ("closed lips") if there are fused clefts in cerebral mantle. In type II ("open lips") the clefts are separated and filled with cerebrospinal fluid connecting lateral ventricle with the subarachnoid space. MATERIAL/Methods: We retrospectively analysed data of patients hospitalized in the Clinical Pediatric Neurology Department of Provincial Hospital No. 2 in Rzeszow between 1998-2011. Clinical data and imaging exams were analysed in the group of children with confirmed schizencephaly. RESULTS: Schizencephaly was recognized in 32 children. Diagnosis was made in children at the ages between 2 weeks and 15 years - the majority of older children were born before the year 2000. Diagnostic imaging, most often magnetic resonance imaging, was performed in all of the children. In most cases coexistence of other CNS malformations was discovered. In only one patient there were no neurological symptoms, most of the children presented different developmental disorders and neurological symptoms - most often cerebral palsy and epilepsy. In the group of children with bilateral and type II schizencephaly certain symptoms occurred more often. Conclusion: Schizencephaly is a rare central nervous

system developmental disorder, which is very often associated with other severe brain malformations and in most of the cases subsequent multiple neurological symptoms. The method of choice in diagnosis of schizencephaly is magnetic resonance, which shows the degree and type of cleft, coexisting abnormalities and allows differential diagnosis. With the increased availability of this method it is possible to recognize schizencephaly more often and earlier.

PMID: 25473439 [PubMed - as supplied by publisher]

55. Res Dev Disabil. 2014 Oct 24;36C:207-212. doi: 10.1016/j.ridd.2014.10.001. [Epub ahead of print]

Estimating the prevalence of cerebral palsy in Taiwan: A comparison of different case definitions.

Chang MJ1, Ma HI2, Lu TH3.

The estimated prevalence of cerebral palsy (CP) worldwide ranged from 0.74 to 3.6 per 1000 live births according to different studies, which may be due to different data sources and case definitions used. We used a representative sample of one million patients (about 1/23 of total population) covered by Taiwan's National Health Insurance (NHI) to estimate the prevalence using different case definitions. Eight years of NHI Research Database claims data for all children born between 1996 and 2000 were reviewed for CP diagnoses. The estimated prevalence of CP (cases per 1000 live births) varied from 4.1 to 1.3 for different case definitions. For a minimum age of 4 years old at diagnosis, a diagnosis made by specialists (pediatricians and physicians of physical medicine and rehabilitation), and the CP diagnosis was mentioned at least three times in claims data, the mean estimated prevalence of CP was 3.2 (95% CI 2.8-3.7). According to this definition, which is most compatible with previous studies, the estimated prevalence in Taiwan was 3.4 (95% CI 2.8-4.0) for boys and 3.1 (95% CI 2.5-3.7) for girls, significantly higher than that in other countries. Additional studies are needed to determine the reasons of higher prevalence in Taiwan.

PMID: 25462481 [PubMed - as supplied by publisher]

55. Zhonghua Yi Xue Yi Chuan Xue Za Zhi. 2014 Dec;31(6):725-9. doi: 10.3760/cma.j.issn.1003-9406.2014.06.009.

A microdeletion of chromosome 9q34.11 may cause suspected cerebral palsy [Article in Chinese]

Li H1, Chen Y, Li Q, Liu M, Mao J, Song D, Li H.

OBJECTIVE: To identify the genetic cause for a child with mental retardation and dyskinesia. Methods: After the routine genetic counseling for the child and the core family members, conventional peripheral blood karyotyping with G-banding and tandem mass spectrometry were applied to find the common genetic problems. Array-comparative genomic hybridization (aCGH) based on the whole genome level was performed to detect minor chromosomal structural abnormalities and the result was confirmed by multiplex ligation dependent probe amplification (MLPA). RESULTS: The proband's karyotype was normal. There were not obvious abnormalities for the testing of 26 types of congenital metabolic diseases. A -2.11 Mb microdeletion of chromosome 9q34.11 region was found though aCGH, which including SPTAN1, TOR1A and other nearly 50 genes related to mental retardation, early infantile spasms, epileptic encephalopathy, myelin dysplasia and dystonia. The -2.11 Mb chromosomal microdeletion was identified by MLPA. Conclusion:
The 2.11 Mb microdeletion of chromosome 9q34.11 region may lead to suspected cerebral palsy. Cytogenetic Methods: combined with MLPA and aCGH can efficiently identify genetic etiology and provide accurate results for clinical diagnosis.

PMID: 25449075 [PubMed - in process]