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Cerebral palsy research news

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Interventions and Management

1. Upper-extremity Spasticity-reducing Treatment in Adjunct to Movement Training and Orthoses in Children with Cerebral Palsy at Gross Motor Function- and Manual Ability Classification System Levels IV-V: A Descriptive Study.
Andersson G, Renström B, Blaszczyk I, Domellöf E.

Dev Neurorehabil. 2019 Aug 22:1-10. doi: 10.1080/17518423.2019.1655677. [Epub ahead of print]

Covering a 20-year period of work with children with severe cerebral palsy (CP) within a Swedish habilitation service, changes in passive wrist extension with fingers extended (PWE-FE) and current hand function are described and compared between children receiving systematic upper-extremity treatment with botulinum neurotoxin type A and intervention programs from before 7 years of age (Group 1, n = 7), those whom for various reasons did not undergo this treatment (Group 2, n = 10), and those not having the option to receive treatment until later during childhood/adolescence (Group 3, n = 8). Group 3 showed more critical and less normal PWE-FE values for both wrists, and poorer hand function scores, particularly compared with Group 1. Findings cautiously suggest that repeated upper-extremity spasticity-reducing treatment and movement training/orthoses from an early age may help prevent critical loss of passive range of motion of the wrist joint flexion/extension and promote hand function development in children with severe CP.

PMID: 31437072

2. Guided Growth Improves Coxa Valga and Hip Subluxation in Children with Cerebral Palsy.
Hsieh HC, Wang TM, Kuo KN, Huang SC, Wu KW.

Clin Orthop Relat Res. 2019 Aug 12. doi: 10.1097/CORR.0000000000000903. [Epub ahead of print]

BACKGROUND: Spastic hip subluxation or dislocation that is associated with an excessive coxa valga deformity is a common pathologic condition in children with cerebral palsy (CP) that is often treated with large bone reconstructive procedures. Guided growth techniques (such as stapling, plate, or transphyseal screw) have been widely used to alter the growth axis in patients with a lower-limb deformity but only a few reports have described their use in patients with coxa valga deformities. QUESTIONS/PURPOSES: (1) Does guided growth surgery using a transphyseal screw combined with adductor tenotomy prevent progressive coxa valga deformity and lateral hip subluxation in children with CP? (2) What factors influence the correction of coxa valga deformity and the success of hip stabilization? (3) What complications were associated with this operation and how often did children treated with it undergo reoperation? METHODS: From 2012 to 2016, at our institution, three authors (H-CH, KNK, K-WW) retrospectively studied data on children with CP who underwent guided growth of the hip for progressive bilateral hip subluxation associated with coxa valga deformities. A single percutaneous screw was inserted across the inferomedial portion of proximal femoral physis in an AP view and centered along femoral neck in lateral view under fluoroscopy guidance. During the period, we treated 25 consecutive children with CP who had progressive hip subluxation with coxa valga deformities. The indications for surgery were migration percentage > 30% and

head-shaft angle > 155° with at least 2 years growth remaining. Of those, 13 patients underwent guided growth alone, and 48% (12) underwent a combination of guided growth and adductor tenotomy. Of the 25 patients treated with this approach, 96% (24) were available for follow-up with complete data at a minimum of 2 years follow-up (mean 50 months; range 25 to 72). All children (17 boys and seven girls; 48 hips) underwent surgery at a mean age of 8 years (range 5 to 12). With regard to the gross motor function classification system, three patients were Level 1, four patients were Level II, seven patients were Level III, seven were Level IV, and three were Level V. Radiographic parameters including the head-shaft angle, Hilgenreiner's epiphyseal angle, acetabular index, and Reimer's migration percentage were assessed before surgery and at the latest follow-up examination by one author (H-CH). Complications and reoperations were assessed by chart review. During the period in question, we generally offered secondary reconstructive surgery to patients who underwent a guided growth procedure once their subluxation progressed. RESULTS: With the data available, the coxa valga and lateral hip subluxation improved in terms of the reduction of head-shaft angle by a mean of 13° ± 7° (95% CI 11 to 15; $p < 0.001$) and the reduction of the migration percentage by 10% ± 11% (95% CI 7 to 13; $p < 0.001$). After controlling for potentially confounding variables like gender, gross motor function classification system, Hilgenreiner's epiphyseal angle and acetabular index, we found that longer follow-up duration ($r = 0.234$; $p < 0.001$) and a smaller preoperative migration percentage ($r = -0.258$; $p = 0.004$) were associated with larger changes in the head-shaft angle. In terms of complications, we found that the proximal femoral physis grew off the screw tip in 44% (21 of 48 hips) at a mean of 28 months. Among these, 31% of hips (15 of 48) in 33% of patients (eight of 24) underwent replacement with a longer screw. Among the 17% of hips (eight of 48) in 21% of patients (five of 24) who had progressive lateral subluxation and underwent secondary reconstructive surgery, we found that their preoperative acetabular index was higher (mean 29° versus 21°; $p < 0.001$), as was their head-shaft angle (mean 166° versus 162°; $p = 0.045$), and migration percentage (mean 54% versus 36 %; $p < 0.001$). CONCLUSIONS: Although guided growth with single transphyseal screw did not create as large a degree of varus as proximal femoral osteotomy, it did stabilize the hip in children with cerebral palsy with migration percentage less than 50% in our series. It is a simple procedure that can be of benefit to children with cerebral palsy with unstable hip. Reoperation in patients where the physis has grown off the screw tip can be a problem; fortunately, it is a rather minor procedure to replace with a longer screw. LEVEL OF EVIDENCE: Level III, therapeutic study.

PMID: 31425278

3. Improvement in gross motor function and muscle tone in children with cerebral palsy related to neonatal icterus: an open-label, uncontrolled clinical trial.
Thanh LN, Trung KN, Duy CV, Van DN, Hoang PN, Phuong ANT, Ngo MD, Thi TN, Viet AB.

BMC Pediatr. 2019 Aug 22;19(1):290. doi: 10.1186/s12887-019-1669-2.

BACKGROUND: Although stem cell transplantation has been successfully performed for cerebral palsy (CP) related to oxygen deprivation, clinical trials involving the use of stem cell transplantation for CP related to neonatal icterus have not been reported. The aim of this study was to evaluate the effectiveness of transplantation of autologous bone marrow mononuclear cell (BMMC) for improving gross motor function and muscle tone in children with CP related to neonatal icterus. METHODS: This open-label, uncontrolled clinical trial, which included 25 patients with CP related to neonatal icterus who had a Gross Motor Function Classification System (GMFCS) score between level II and level V, was conducted between July 2014 and July 2017 at Vinmec International Hospital (Vietnam). BMMC were harvested from the patients' iliac crests. Two procedures involving BMMC transplantation via the intrathecal route were performed: the first transplantation was performed at baseline, and the second transplantation was performed 6 months after the first transplantation. Gross motor function and muscle tone were measured at three time points (baseline, 6 months, and 12 months) using the Gross Motor Function Measure (GMFM) and the Modified Ashworth Scale. RESULTS: In this trial, we observed significant improvement in gross motor function and a significant decrease in muscle tone values. Total score on the 88-item GMFM (GMFM-88), scores on each GMFM-88 domain, and the 66-item GMFM (GMFM-66) percentile were significantly enhanced at 6 months and 12 months after the first transplantation compared with the corresponding baseline measurements (p-values < 0.05). In addition, a significant reduction was observed in muscle tone score after the transplantations (p-value < 0.05). CONCLUSION: Autologous BMMC transplantation can improve gross motor function and muscle tone in children with CP related to neonatal icterus. TRIAL REGISTRATION: ClinicalTrials.gov identifier: NCT03123562 . Retrospectively registered on December 26, 2017.

PMID: 31438885

4. Biodex balance training versus conventional balance training for children with spastic diplegia.
El-Gohary TM, Emara HA, Al-Shenqiti A, Hegazy FA.

J Taibah Univ Med Sci. 2017 Aug 18;12(6):534-540. doi: 10.1016/j.jtumed.2017.07.002. eCollection 2017 Dec.

OBJECTIVE: The purpose of this study was to compare the effectiveness of balance training using the Biodex balance system

and a conventional balance training programme on balance score and on gross motor skills of children with spastic diplegia. METHODS: A randomized controlled study was conducted on 48 spastic diplegic children with cerebral palsy (26 boys and 22 girls) in the age range of 5-8 years. The children were randomly allocated to two equal groups. The investigators performed balance and gross motor function assessments for every child using the paediatric Berg balance scale and the gross motor function measure -88 scale (dimensions D and E) before and after the treatment programme. Passive repositioning sense was measured by a Biodex III isokinetic dynamometer. The study group received Biodex balance training and traditional physical therapy programme training, whereas the control group received conventional balance training in addition to the traditional physical therapy programme training, 3 times per week for 12 weeks. RESULTS: Significant improvement was observed in all outcome measures of the two groups, comparing their pre- and post-treatment mean values. Furthermore, the results revealed a significant ($P < 0.05$) improvement in mean post-treatment values for the Biodex balance training group. CONCLUSION: Balance training using the Biodex balance system is superior to conventional balance training for improving the balance abilities and gross motor functions of children with cerebral palsy and spastic diplegia.

PMID: 31435291

5. Effects of forward tilted seating and foot-support on postural adjustments in children with spastic cerebral palsy: An EMG-study.
Angsupaisal M, Dijkstra LJ, la Bastide-van Gemert S, van Hoorn JF, Burger K, Maathuis CGB, Hadders-Algra M.

Eur J Paediatr Neurol. 2019 Aug 1. pii: S1090-3798(19)30016-9. doi: 10.1016/j.ejpn.2019.07.001. [Epub ahead of print]

OBJECTIVE: To evaluate the effect of 15° forward (FW) seat inclination and foot-support in children with cerebral palsy (CP) on postural adjustments during reaching. DESIGN: Observational study repeated-measures design; step two of two-step-project. SETTING: Laboratory unit within University Hospital and two special education schools. PARTICIPANTS: 19 children (ten unilateral spastic CP (US-CP); nine bilateral spastic CP (BS-CP); Gross Motor Function Classification System levels I-III; 6-12 years old). Participants were able to take part for one one-hour session. INTERVENTION: Reaching while sitting in four seating conditions (FW or horizontal seat; with or without foot-support) applied in randomized order. OUTCOME MEASURES: Simultaneously, surface electromyography (EMG) of neck, trunk and arm muscles and kinematics of head and reaching arm (step one of two-step-project) were recorded. Primary outcome parameters were the ability to modulate EMG-amplitudes at baseline and during reaching (phasic muscle activity). Other EMG-parameters were direction-specificity (1st control level), and 2nd level of control parameters: recruitment order, and anticipatory postural activity. Motor behaviour measures: ability to modulate EMG-amplitudes to kinematic characteristics of reaching and head stability. RESULTS: Only foot-support was associated with increased tonic background EMG-amplitudes and decreased phasic EMG-amplitudes of the trunk extensors in children with US-CP and BS-CP (mixed-models analyses; p-values <0.01). The foot-support effect was also associated with better kinematics of reaching (Spearman's Rho; p-values <0.01). CONCLUSION: In terms of postural adjustments during forward reaching, foot-support enhanced the children's capacity to modulate trunk extensor activity, which was associated with improved reaching quality. FW-tilting did not affect postural muscle activity.

PMID: 31420131

6. Effectiveness of Lower-Extremity Functional Training (LIFT) in Young Children With Unilateral Spastic Cerebral Palsy: A Randomized Controlled Trial.
Surana BK, Ferre CL, Dew AP, Brandao M, Gordon AM, Moreau NG.

Neurorehabil Neural Repair. 2019 Aug 22:1545968319868719. doi: 10.1177/1545968319868719. [Epub ahead of print]

Background. Children with unilateral spastic cerebral palsy (USCP) have strength, coordination, and balance deficits affecting gross motor skills, such as walking, running, and jumping. However, there is a paucity of evidence for effective treatments for lower-extremity (LE) function in children with USCP. Objective. To determine the effectiveness of LE intensive functional training (LIFT) compared with an attention control group receiving upper-extremity bimanual training (Hand-Arm Bimanual Intensive Therapy [H-HABIT]). Methods. A total of 24 children with USCP were randomized to receive 90 hours of LIFT (5.8 [2.3] years) or an equivalent dosage of H-HABIT (5.1 [2.6] years) delivered 2 h/d, 5 d/wk for 9 weeks. Caregivers were trained to administer the intervention in the home setting. Progress and skill progression were monitored, and supervision was provided via weekly telerehabilitation. The primary outcome was the 1-minute walk test (1MWT). Secondary outcomes included self-selected and fast walking speeds, ABILOCO-kids, 30-s chair rise test, and single-leg stance. Results. LIFT showed greater improvement for the 1MWT ($P = .017$) and ABILOCO-kids ($P = .008$) compared with controls. The other secondary outcomes were not different between groups. Conclusions. The administration of LE intensive interventions in the home setting by caregivers was shown to be an effective and novel mode of delivery for improving gait capacity and performance. LIFT delivered in the home setting using telerehabilitation for monitoring resulted in improvements in ambulation distance and overall walking ability as compared to an intervention of equal intensity and duration that also

controlled for the increased social interaction and attention between caregiver and child.

PMID: 31434537

7. Comparison of two different methods in preoperative planning of the amount of lengthening in Z achilloplasty technique.
Ozyalvac ON, Akpinar E.

J Pediatr Orthop B. 2019 Aug 16. doi: 10.1097/BPB.0000000000000666. [Epub ahead of print]

Achilles tendon lengthening (ATL) surgery is a technique that is frequently used in the surgical treatment of contracture of the Achilles tendon seen in many pediatric orthopedic problems such as cerebral palsy, clubfoot, pes planovalgus and myelomeningocele. It is important to appropriately adjust the amount of ATL. However, the literature on the preoperative calculation of the required amount of tendon lengthening is limited. The aim of the study was to compare the reliability of the two different methods of predicting the amount of ATL. Eighteen feet of 16 patients who underwent ATL with Z-plasty technique were included in the study. The required amount of ATL was calculated as double blind according to the Cosine theorem and a method that was described by Garbarino et al. in 1985 and compared with the amounts of ATL applied during the operation. The mean amount of lengthening was 25.24 mm during surgery. The required amount of lengthening was 41.55 ± 11.0 mm, according to the Garbarino's method. The required amount of lengthening was 23.93 ± 9.03 mm, according to the Cosine theorem. The quantities calculated according to the Cosine theorem showed excellent agreement with the amount of lengthening during surgery. The quantities calculated according to the Garbarino's method showed poor agreement with the amount of lengthening during surgery. The calculation of the amount of ATL required in the treatment of the equinus deformity before surgery is possible by Cosine Theorem. The method of Cosine theorem is more reliable than the previous method described by Garbarino et al.

PMID: 31425334

8. Performance of school-aged children with cerebral palsy at GMFCS levels I and II on high-level, sports-focussed gross motor assessments.
Clutterbuck GL, Auld ML, Johnston LM.

Disabil Rehabil. 2019 Aug 21:1-9. doi: 10.1080/09638288.2019.1650964. [Epub ahead of print]

Purpose: To investigate performance of children with cerebral palsy (CP) at GMFCS Levels I-II on sports-focussed gross motor assessments; and concurrent validity of mainstream and CP-specific high-level assessment batteries and field tests. Methods: Fifty-four children (6-12 years) with CP at GMFCS I-II completed the Test of Gross Motor Development-second edition, Gross Motor Function Measure-Challenge Module, Muscle Power Sprint Test (MPST), 10 × 5 m Sprint Test (10 × 5 mST), Vertical Jump, Broad Jump, and Seated Throw. Correlations between measures, age and mobility level, and group differences between age and mobility level were examined and content analysis performed. Results: Children at GMFCS I demonstrated significantly higher gross motor assessment battery scores than children at GMFCS II ($U = 73.5$-109.0, $p < 0.001$). Performance improved with age for children at Level I but not II. Children with higher overall motor scores scored higher on running (MPST, 10 × 5 mST, $r = -0.516$ to -0.816, $p < 0.001$), jumping (Vertical Jump, Broad Jump, $r = 0.499$-0.774, $p < 0.001$) and throwing (Seated Throw, $r = 0.341$-0.500, $p = 0.012 < 0.001$) field tests. Conclusions: High-level gross motor assessments were achievable and appropriately challenging for children with CP at GMFCS I-II. Scores discriminated between performance and were associated with mobility level. Concurrent validity was established between gross motor assessment batteries, and locomotor field tests. IMPLICATIONS FOR REHABILITATION For children with cerebral palsy at GMFCS I-II, sports-focussed assessments should be used to assess high-level gross motor function. The Gross Motor Function Measure-Challenge and Test of Gross Motor Development demonstrate no ceiling for children with cerebral palsy at GMFCS I-II. Single-item running and jumping field tests provide targeted skill assessment and estimate sports skills for children with cerebral palsy.

PMID: 31433244

9. Effectiveness of surf therapy for children with disabilities.
Clapham ED, Lamont LS, Shim M, Lateef S, Armitano CN.

Disabil Health J. 2019 Aug 2:100828. doi: 10.1016/j.dhjo.2019.100828. [Epub ahead of print]

BACKGROUND: Few researchers have examined the effects of surf programs on children with disabilities. Due to previous research findings, surfing is being used, as the focus of physical activity intervention due to its numerous health and therapeutic benefits. OBJECTIVE/HYPOTHESIS: The purpose of this study was to explore the effects of an eight-week surfing intervention on various physical fitness measures in 71 children with disabilities such as autism spectrum disorder, down syndrome, global developmental delays, and cerebral palsy. The study also sought to compare the differences in overall fitness levels between the surf therapy group and an unstructured pool playgroup. Researchers predicted significant differences in the surf therapy group. METHODS: The assessment procedure consisted of pre and post physical fitness measures selected from the Brockport Physical Fitness Test in two groups: surfing (n = 71) and an unstructured aquatic program (n = 20). RESULTS: The results demonstrated significant improvements in core strength (p = 0.00), upper body strength (p = 0.00), flexibility (p = 0.01) and cardiorespiratory endurance (p = 0.00) in the surfing group. However, there were no significant differences in overall fitness levels between the surfing and unstructured pool playgroups. Body composition measurements on the surfing group demonstrated a significant reduction in total body fat % (p = 0.016) and fat free mass (p = 0.008) and a significant improvement in bone mineral density (p = 0.004) pre to post surf therapy. CONCLUSIONS: This research demonstrated the effectiveness and physiological benefits of surf therapy for children with selected disabilities.

PMID: 31422168

10. Automated movement analysis to predict motor impairment in preterm infants: a retrospective study.
Raghuram K, Orlandi S, Shah V, Chau T, Luther M, Banihani R, Church P.

J Perinatol. 2019 Aug 20. doi: 10.1038/s41372-019-0464-0. [Epub ahead of print]

OBJECTIVE: To apply automated movement analysis to the general movements assessment (GMA) to build a predictive model for motor impairment (MI). STUDY DESIGN: A retrospective cohort study including infants ≤306/7 weeks GA or BW ≤1500 g seen at 3-5 months was conducted. Automated video analysis was used to develop a multivariable model to identify MI, defined as Bayley motor composite score <85 or cerebral palsy (CP). RESULTS: One hundred and fifty two videos were analyzed. Median GA and BW were 275/7 weeks and 955 g, respectively. MI and CP rates were 22% (N = 33) and 14% (N = 22). Minimum, mean, and mean vertical velocity of the infant's silhouette correlated significantly with MI. Sensitivity, specificity, positive and negative predictive values, and accuracy of automated GMA were 79%, 63%, 37%, 91%, and 66%, respectively. C-statistic indicated good fit (C = 0.77). CONCLUSIONS: Automated movement analysis predicts MI in preterm infants. Further refinement of this technology is required for clinical application.

PMID: 31431653

11. As-indicated versus routine vision screening of preterm children: a 17-year retrospective regional study.
Welinder L, Bender L, Eriksen HH, Nissen KR, Ebbesen F.

Acta Ophthalmol. 2019 Aug 17. doi: 10.1111/aos.14227. [Epub ahead of print]

PURPOSE: To investigate outcomes of routine vision screening compared to as-indicated ophthalmological investigation of all children born preterm in a Danish region from 1997 to 2014. METHODS: All children born preterm (gestation age < 32 weeks or birthweight < 1500 g) screened for retinopathy of prematurity (ROP) were divided into two groups. From 1997 to 2009, only children treated for ROP or referred for visual problems received ophthalmological investigation (as-indicated group). From 2010 to 2014, all ROP-screened infants were offered ophthalmological investigation at 6 months and 3 years of age (screening group). RESULTS: A total of 560 children were included in the as-indicated period, 41 and 87 were referred for ophthalmological investigation at 6 months and 3 years, respectively. In the screening period, 295 children were included, 251 and 150 of whom underwent vision evaluation at 6 months and 3 years, respectively. Mean visual acuity was 4.1 cycles per degree with Teller acuity cards at 6 months and 0.78 decimal at 3 years. At 3 years, 2.7%(n = 11) in the as-indicated versus 3.5%(n = 10) screening group had visual acuity < 6/18 (p = 0.24). Cerebral palsy (n = 28) and epilepsy (n = 5) were significantly related to vision impairment (p = 0.001/0.006), while treated ROP was not (n = 13). Refractive error was common at 3 years (61%), especially astigmatism (50%). Gestational age, birthweight and ROP were not associated with vision impairment or refractive error. CONCLUSION: Screening preterm children at 6 months and 3 years did not reveal more visually impaired children compared to examination when indicated.

PMID: 31421027

12. Laparoscopic fundoplication for a child with abdominal intrathecal Baclofen pump.
Okata Y, Bitoh Y, Aida Y, Miyauchi H, Nakatani T, Tomioka Y.

Asian J Endosc Surg. 2019 Aug 20. doi: 10.1111/ases.12752. [Epub ahead of print]

This is the first case report describing a laparoscopic fundoplication in a child with an intrathecal Baclofen pump which was inserted because of severe spasticity secondary to cerebral palsy. The child had symptoms of gastroesophageal reflux with recurrent episodes of aspiration pneumonia. These were managed with a gastrostomy and conservative therapy with no success. The presence of an intrathecal Baclofen pump makes abdominal surgery challenging and carries the risk of pump infection with its associated sequelae. However, we performed a successful laparoscopic fundoplication with no intraoperative complications and the child was asymptomatic at 18 months follow-up.

PMID: 31430037

13. Is Loop Ileostomy in Patients with Cecal Bascule a Viable Option?
Shetty T, Ivanics T, Nasser H, Stefanou A.

Case Rep Surg. 2019 Jul 22;2019:8549692. doi: 10.1155/2019/8549692. eCollection 2019.

BACKGROUND: Cecal bascule, initially described in 1899 by Treves, is the rarest form of cecal volvulus and represents a phenomenon when a redundant and distended cecum folds anteriorly over the ascending colon causing an intestinal obstruction. Patients with cerebral palsy are at increased risk for this condition. CASE PRESENTATION: We present a 28-year-old male with cerebral palsy, functionally dependent in all activities of daily living, who had undergone a loop ileostomy for cecal bascule. He then presented to our emergency department with a large loop ileostomy prolapse, which was the result of an inverted prolapsed cecum through the efferent ileostomy limb. He underwent a right hemicolectomy with end ileostomy and transverse mucous fistula creation through the previous ostomy site. He progressed well appropriately postoperatively and was discharged home. CONCLUSIONS: While cecal bascule is a rare form of bowel obstruction, patients with cerebral palsy are at an increased risk for this condition. The treatment options are numerous and are primarily surgical. Diverting loop ileostomy alone is not a recommended treatment. A high index of suspicion is warranted in all cases of large bowel obstruction to minimize risk of recurrence, morbidity, and mortality for patients afflicted by this condition.

PMID: 31428508

14. Craniopharyngiomas presenting as incidentalomas: results of KRANIOPHARYNGEOM 2007.
Boekhoff S, Bison B, Eveslage M, Sowithayasakul P, Müller HL.

Pituitary. 2019 Aug 22. doi: 10.1007/s11102-019-00983-7. [Epub ahead of print]

PURPOSE: Childhood-onset craniopharyngiomas (CP) are diagnosed due to clinical symptoms (symCP) or incidentally (incCP). We investigated clinical manifestations and outcome in incCPs and symCPs. METHODS: IncCP were discovered in 4 (3 m/1 f) and symCP in 214 (101 m/113 f) CP recruited 2007-2014 in KRANIOPHARYNGEOM 2007. Age, sex, height, body mass index (BMI), tumor volume, degree of resection, pre- and postsurgical hypothalamic involvement/lesions, pituitary function and outcome were compared between both subgroups. RESULTS: Reasons for imaging in incCP were cerebral palsy, head trauma, nasal obstruction, and tethered-cord syndrome, whereas headache (44%), visual impairment (25%), and growth retardation (17%) lead to imaging in symCP. Tumor volume at diagnosis was smaller in incCP (median 2.39 cm3; range 0.14-4.10 cm3) when compared with symCP (15.86 cm3; 0.002-286.34 cm3). Age, gender, BMI, height, hydrocephalus, tumor location, and hypothalamic involvement at diagnosis of incCP were within the range of these parameters in symCP. Complete resections were achieved more frequently (3/4 patients) in incCP when compared with symCP (20%). Surgical hypothalamic lesions were distributed similar in incCP and symCP. Irradiation was performed only in symCP (33%). No noticeable differences were observed concerning survival rates, endocrine deficiencies, BMI, height, functional capacity and quality of life of the 4 incCP cases when compared with the symCP cohort. CONCLUSIONS: IncCP are rare (1.8%) and characterized by lack of endocrine deficiencies, resulting in normal height and BMI, no hydrocephalus, and smaller tumor volume at diagnosis when compared with symCPs. Outcome of the observed incCP is similar with symCP. CLINICAL TRIAL REGISTRATION NUMBER: NCT01272622.

PMID: 31440945

15. High risk of long-term impairment in donor twins with spontaneous twin anemia polycythemia sequence.
Tollenaar LSA, Lopriore E, Slaghekke F, Oepkes D, Middeldorp JM, Haak MC, Klumper FJCM, Tan RNGB, Rijken M, Van Klink JMM.

Ultrasound Obstet Gynecol. 2019 Aug 20. doi: 10.1002/uog.20846. [Epub ahead of print]

OBJECTIVE: The aim of this study was to evaluate the long-term neurodevelopmental and behavioural outcome in survivors of twin anemia polycythemia sequence (TAPS). METHODS: Neurological, motor, cognitive, and behavioural development was assessed in a consecutive cohort of spontaneous TAPS survivors born between 2005 and 2017 at the Leiden University Medical Centre, The Netherlands. The primary outcome was neurodevelopmental impairment (NDI), a composite outcome including: cerebral palsy, deafness, blindness and motor and/or cognitive delay. NDI was subdivided into two grades of severity: mild-to-moderate and severe NDI. RESULTS: In total, 49 twin pregnancies complicated by spontaneous TAPS were included in the study. Perinatal survival was 83% (81/98). Neurodevelopmental assessment was performed in 91% (74/81). NDI occurred in 31% (22/74) of the TAPS survivors, and was found more often in donors (44%; 15/34) than in recipients (18%; 7/40) (OR 4.1, 95%CI 1.6-8.6, p = 0.002). Severe NDI was detected in 9% (7/74) and was higher in donors compared to recipients, 18% (6/34) versus 3% (1/40), respectively (p = 0.056). Donors demonstrated lower cognitive scores compared to their recipient co-twins (p = 0.011). Bilateral deafness was detected in 15% (5/34) of the donors versus 0% (0/40) in recipients (p = 0.056). Parents reported to have more developmental concerns about their donor twin than their recipient twin (p = 0.001). Risk factors for NDI were gestational age at birth (OR = 0.7, 95%CI 0.5-0.9, p = 0.003) and severe anaemia (OR = 6.4, 95%CI 2.4-17.0, P < 0.001). CONCLUSION: TAPS donors have a fourfold higher risk of NDI compared to their recipient co-twins and are at increased risk of cognitive delay and deafness. This article is protected by copyright. All rights reserved.

PMID: 31432580

16. Hospital survey showed wide variations in therapeutic hypothermia for neonates in Germany.
Giannakis S, Ruhfus M, Rüdiger M, Sabir H; German Neonatal Hypothermia Network.

Acta Paediatr. 2019 Aug 20. doi: 10.1111/apa.14979. [Epub ahead of print]

Perinatal asphyxia is a major contributor to neonatal death in term born infants. Hypoxic-ischaemic encephalopathy (HIE) is the most important asphyxia-related complication affecting the newborn brain and causes 23% of neonatal deaths worldwide. Severe disabilities, such as cerebral palsy, seizures or mental retardation, occur in 30% of newborn infants with moderate HIE and 90% with severe HIE. Therapeutic hypothermia is the only established, standardised treatment and its efficacy has been proven by many randomised clinical trials. This article is protected by copyright. All rights reserved.

PMID: 31432551

17. Inequity in physiotherapeutic interventions for children with Cerebral Palsy in Sweden - a national registry study.
Degerstedt F, Enberg B, Keisu BI, Björklund M.

Acta Paediatr. 2019 Aug 22. doi: 10.1111/apa.14980. [Epub ahead of print]

AIM: The aim of this study was to investigate the distribution of physiotherapeutic interventions for children with Cerebral Palsy in Sweden from an equity perspective, considering sex, country of birth and geographical region. METHOD: This national cross-sectional registry study includes children with Cerebral Palsy aged 0-18 years who participated in 2015 in the Swedish national quality registry, the Cerebral Palsy follow-up program, CPUP. Comparisons and associations between physiotherapeutic interventions and sex, country of birth and geographical regions were conducted using Chi2 and logistic regression analysis, controlling for cognitive level, level of motor function, age group and dominating symptom. RESULTS: Of the 2855 participants, 2201 (79%) had received physiotherapy. Children born in Sweden had 1.60 times higher odds (95% CI 1.10-2.33) of receiving physiotherapy compared with children born in foreign countries. Distribution of physiotherapeutic interventions differed significantly between geographical regions. No associations between sex and physiotherapeutic interventions were observed. CONCLUSION: The results of this study indicate inequity in care in Sweden towards children with Cerebral Palsy born in other counties. Further, physiotherapeutic interventions were not equally distributed in different geographical regions of Sweden. Knowledge of inequity is crucial in order to address the problem. This article is protected by copyright. All rights reserved.

PMID: 31435959

18. From congenial paralysis to post-early brain injury developmental condition: Where does cerebral palsy actually stand?
Chabrier S, Pouyfaucon M, Chatelin A, Bleyenheuft Y, Fluss J, Gautheron V, Newman CJ, Sébire G, Bogaert PV, Vuillerot C, Brochard S, Dinomais M.

Ann Phys Rehabil Med. 2019 Aug 14. pii: S1877-0657(19)30113-7. doi: 10.1016/j.rehab.2019.07.003. [Epub ahead of print]

Cerebral palsy (CP), an umbrella term for a developmental motor disorder caused by early brain injury (EBI)/interference, remains debated. In this essay, we present a narrative, beginning with the original anatomical-clinical description of the so-called paralysie congéniale (congenial paralysis) by the French psychiatrist Jean-Baptiste Cazauvieilh. We then discuss how the concept has evolved over the last 2 centuries. We aim to illustrate these ideas with the biopsychosocial model of health, especially in light of the current neuroscientific and sociological knowledge of human development. We endeavour to integrate 3 connected but distinct entities: 1) the EBI as a seminal turning point of the individual's story, 2) the clinical findings we call CP, when motor impairment and activity limitation related to post-EBI (or other early non-progressive brain interference) appears, and 3) a post-EBI developmental condition that encompasses the overall consequences of an EBI. This framework should guide individual, familial and collective care discussions and research strategies beyond the scope of CP.

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Prevention and Cure

19. Topical Administration of a Soluble TNF Inhibitor Reduces Infarct Volume After Focal Cerebral Ischemia in Mice.
Yli-Karjanmaa M, Clausen BH, Degn M, Novrup HG, Ellman DG, Toft-Jensen P, Szymkowski DE, Stensballe A, Meyer M, Brambilla R, Lambertsen KL.

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BACKGROUND: Tumor necrosis factor, which exists both as a soluble (solTNF) and a transmembrane (tmTNF) protein, plays an important role in post-stroke inflammation. The objective of the present study was to test the effect of topical versus intracerebroventricular administration of XPro1595 (a solTNF inhibitor) and etanercept (a solTNF and tmTNF inhibitor) compared to saline on output measures such as infarct volume and post-stroke inflammation in mice. METHODS: Adult male C57BL/6 mice were treated topically (2.5 mg/ml/1μl/h for 3 consecutive days) or intracerebroventricularly (1.25 mg/kg/0.5 ml, once) with saline, XPro1595, or etanercept immediately after permanent middle cerebral artery occlusion (pMCAO). Mice were allowed to survive 1 or 3 days. Infarct volume, microglial and leukocyte profiles, and inflammatory markers were evaluated. RESULTS: We found that topical, and not intracerebroventricular, administration of XPro1595 reduced infarct volume at both 1 and 3 days after pMCAO. Etanercept showed no effect. We observed no changes in microglial or leukocyte populations. XPro1595 increased gene expression of P2ry12 at 1 day and Trem2 at 1 and 3 days, while decreasing Cx3cr1 expression at 1 and 3 days after pMCAO, suggesting a change in microglial activation toward a phagocytic phenotype. CONCLUSION: Our data demonstrate that topical administration of XPro1595 for 3 consecutive days decreases infarct volumes after ischemic stroke, while modifying microglial activation and the inflammatory response post-stroke. This suggests that inhibitors of solTNF hold great promise for future neuroprotective treatment in ischemic stroke.

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