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Professor Nadia Badawi

Macquarie Group Foundation Chair of Cerebral Palsy
PO Box 560, Darlinghurst, New South Wales 2010 Australia

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Interventions and Management

1. J Hand Surg Am. 2014 Mar 6. pii: S0363-5023(14)00137-3. doi: 10.1016/j.jhsa.2014.01.039. [Epub ahead of print]

Treatment of Swan Neck Deformity in Cerebral Palsy.

Carlson EJ1, Carlson MG2.

Swan neck deformity in patients with cerebral palsy can result from hand intrinsic muscle spasticity or overpull of the digital extensors. After accurate identification of the etiology of the deformity, surgical treatment is directed at correcting the underlying muscle imbalance. Intrinsic lengthening can be used to treat intrinsic muscle spasticity, whereas central slip tenotomy is employed when digital extensor overpull is the deforming force. Accurate diagnosis and application of the proper surgical technique are essential when treating swan neck deformity in patients with cerebral palsy.

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[PMID: 24613587](https://pubmed.ncbi.nlm.nih.gov/24613587/) [PubMed - as supplied by publisher]

2. Exp Brain Res. 2014 Mar 13. [Epub ahead of print]

Improvements in hand function after intensive bimanual training are not associated with corticospinal tract dysgenesis in children with unilateral cerebral palsy.

Friel KM1, Kuo HC, Carmel JB, Rowny SB, Gordon AM.

Unilateral cerebral palsy (CP) results from damage to the developing brain that occurs within the first 2 years of life. Previous studies found associations between asymmetry in the size of the corticospinal tract (CST) from the two hemispheres and severity of hand impairments in children with unilateral CP. The extent to which CST damage affects the capacity for hand function improvement is unknown. This study examines the association between an estimate of CST dysgenesis and (1) hand function and (2) the efficacy of intensive bimanual training in improving hand function. Children with unilateral CP, age 3.6-14.9 years, n = 35, received intensive bimanual training. Children engaged in bimanual functional/play activities (6 h/day, 15 days). Peduncle asymmetry, an estimate of CST dysgenesis, was measured on T1-weighted magnetic resonance imaging scans. Hand function was measured pre- and post-treatment using the assisting hand assessment (AHA) and Jebsen-Taylor test of

hand function (JTTHF). AHA and JTTHF improved post-treatment ($p < 0.001$). Peduncle asymmetry was correlated with baseline AHA and JTTHF ($p < 0.001$) but not with AHA or JTTHF improvement post-training ($R^2 < 0.1$, $p > 0.2$). An estimate of CST dysgenesis is correlated with baseline hand function but is a poor predictor of training efficacy, possibly indicating a flexibility of developing motor systems to mediate recovery.

[PMID: 24623352](#) [PubMed - as supplied by publisher]

3. *Dev Med Child Neurol.* 2014 Mar 12. doi: 10.1111/dmcn.12428. [Epub ahead of print]

Clinical and research considerations in using the Melbourne Assessment 2.

Wallen M.

Author information

[PMID: 24617648](#) [PubMed - as supplied by publisher]

4. *Hosp Pediatr.* 2013 Jul;3(3):233-41.

Outcomes and costs associated with hospitalist co-management of medically complex children undergoing spinal fusion surgery.

Rappaport DI1, Adelizzi-Delany J, Rogers KJ, Jones CE, Petrini ME, Chaplinski K, Ostasewski P, Sharif I, Pressel DM.

OBJECTIVE: The goal of this study was to assess outcomes and costs associated with hospitalist comanagement of medically complex children undergoing spinal fusion surgery for neuromuscular scoliosis. **METHODS:** A hospitalist comanagement program was implemented at a children's hospital. We conducted a retrospective case series study of patients during 2003-2008 to compare clinical and cost outcomes for 87 preimplementation patients, 40 patients during a partially implemented program, and 80 patients during a fully implemented program.

RESULTS: When compared with preimplementation patients, full implementation program patients did not demonstrate a statistically significant difference in median length of stay on the medical/surgical unit after transfer from the PICU (median: 6 vs 8 days; $P = .07$). Patients in the full implementation group received fewer days of parenteral nutrition (median: 0 vs 6 days; $P = .0006$) and had fewer planned and unplanned laboratory studies on the inpatient unit. There was no statistically significant change in returns to the operating room ($P = .08$ between preimplementation and full implementation), other complications, or 30-day readmissions. Median hospital costs increased from preimplementation (\$59372) to partial implementation (\$89302) and remained elevated during full implementation (\$81 651) compared with preimplementation ($P = .004$). Mean physician costs followed a similar trajectory from preimplementation (\$18425) to partial implementation (\$24101) to full implementation (\$22578; $P = .0006$ [versus preimplementation]). **CONCLUSIONS:** A hospitalist comanagement program can significantly affect the care of medically complex children undergoing spinal fusion surgery. Initial program costs may increase.

Comment in: How best to design surgical comanagement services for pediatric surgical patients? [*Hosp Pediatr.* 2013]

[PMID: 24313092](#) [PubMed - indexed for MEDLINE]

5. *J Pediatr.* 2014 Mar 4. pii: S0022-3476(14)00057-2. doi: 10.1016/j.jpeds.2014.01.029. [Epub ahead of print]

Population Pharmacokinetics of Oral Baclofen in Pediatric Patients with Cerebral Palsy.

He Y1, Brunstrom-Hernandez JE2, Thio LL2, Lackey S3, Gaebler-Spira D4, Kuroda MM5, Stashinko E6, Hoon AH Jr6, Vargus-Adams J7, Stevenson RD8, Lowenhaupt S9, McLaughlin JF10, Christensen A10, Dosa NP11, Butler M12, Schwabe A13, Lopez C14, Roge D15, Kennedy D15, Tilton A16, Krach LE17, Lewandowski A18, Dai H19, Gaedigk A20, Leeder JS20, Jusko WJ21.

OBJECTIVE: To characterize the population pharmacokinetics (PK) of oral baclofen and assess impact of patient-

specific covariates in children with cerebral palsy (CP) in order to support its clinical use. **SUBJECTS DESIGN:** Children (2-17 years of age) with CP received a dose of titrated oral baclofen from 2.5 mg 3 times a day to a maximum tolerated dose of up to 20 mg 4 times a day. PK sampling followed titration of 10-12 weeks. Serial R- and S-baclofen plasma concentrations were measured for up to 16 hours in 49 subjects. Population PK modeling was performed using NONMEM 7.1 (ICON PLC; Ellicott City, Maryland). **RESULTS:** R- and S-baclofen showed identical concentration-time profiles. Both baclofen enantiomers exhibited linear and dose/kg-proportional PK, and no sex differences were observed. Average baclofen terminal half-life was 4.5 hours. A 2-compartment PK model with linear elimination and transit absorption steps adequately described concentration-time profiles of both baclofen enantiomers. The mean population estimate of apparent clearance/F was 0.273 L/h/kg with 33.4% inter-individual variability (IIV), and the apparent volume of distribution (Vss/F) was 1.16 L/kg with 43.9% IIV. Delayed absorption was expressed by a mean transit time of 0.389 hours with 83.7% IIV. Body weight, a possible genetic factor, and age were determinants of apparent clearance in these children. **CONCLUSION:** The PK of oral baclofen exhibited dose-proportionality and were adequately described by a 2-compartment model. Our population PK findings suggest that baclofen dosage can be based on body weight (2 mg/kg per day) and the current baclofen dose escalation strategy is appropriate in the treatment of children with CP older than 2 years of age.

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6. Laser Ther. 2012 Mar 28;21(1):23-31. doi: 10.5978/islsm.12-OR-05.

Bone Metabolism in Cerebral Palsy and the Effect of Light-Emitting Diode (LED) Irradiation.

Asagai Y1, Yamamoto K2, Ohshiro T3, Ohshiro T4.

In recent years, through the availability of examination by bone metabolism markers, diagnosis and treatment for osteoporosis in elderly people has been greatly advanced. However, bone metabolism in cases of cerebral palsy has not been fully examined. Though children with cerebral palsy tend to be susceptible to insufficiency fractures, a method of treatment for insufficiency fractures has not been established. In the longitudinal progress of bone metabolism, although there was a difference depending on the severity, reduced bone resorption tended to be mild but osteo-genesis tended to decrease in the severe cases. Osteogenesis and bone resorption markers decreased at around ages 8 and 15. The bone resorption marker maintained mild advancement after age 15. With LED irradiation, all of IGF-1, ucOC, osteogenic marker; BAP, and urinary bone resorption marker; NTx/Cr showed a tendency to normalize. In particular, IGF-1, BAP, and NTx/Cr increased significantly one month after irradiation, compared to the non-irradiation group. Bone density assessed by the DIP method showed no apparent change in the short term either. Irradiation by a commercial LED light bulb indicated a possible positive effect on bone metabolism for children with severe cerebral palsy.

[PMID: 24610978](#) [PubMed]

7. J Spinal Cord Med. 2014 Jan 21. [Epub ahead of print]

Functional electrical stimulation cycling in youth with spinal cord injury: A review of intervention studies.

Mayson TA, Harris SR.

Context Preliminary research suggests that functional electrical stimulation cycling (FESC) might be a promising intervention for youth with spinal cord injury (SCI). Objective To review the evidence on FESC intervention in youth with SCI. Methods Systematic literature searches were conducted during December 2012. Two reviewers independently selected titles, abstracts, and full-text articles. Of 40 titles retrieved, six intervention studies met inclusion criteria and were assessed using American Academy for Cerebral Palsy and Developmental Medicine Levels of Evidence and Conduct Questions for Group Design. Results The study results were tabulated based on levels of evidence, with outcomes categorized according to the International Classification of Functioning, Disability, and Health framework. Evidence from the six included studies suggests that FESC is safe for youth with SCI, with no increase in knee/hip injury or hip displacement. Results from one level II randomized controlled trial suggest that a thrice weekly, 6-month FESC program can positively influence VO₂ levels when compared with passive cycling, as well as quadriceps strength when compared with electrical stimulation and passive cycling. Conclusions FESC

demonstrates limited yet encouraging results as a safe modality to mitigate effects of inactivity in youth with SCI. More rigorous research involving a greater number of participants is needed before clinicians can be confident of its effectiveness.

[PMID: 24621033](#) [PubMed - as supplied by publisher]

8. Clin Rehabil. 2014 Mar 10. [Epub ahead of print]

Exercise training programs to improve hand rim wheelchair propulsion capacity: a systematic review.

Zwinkels M1, Verschuren O, Janssen TW, Ketelaar M, Takken T; on behalf of the Sport-2-Stay-Fit study group. Collaborators (9)

Objective: An adequate wheelchair propulsion capacity is required to perform daily life activities. Exercise training may be effective to gain or improve wheelchair propulsion capacity. This review investigates whether different types of exercise training programs are effective in improving wheelchair propulsion capacity. **Data sources:** PubMed and EMBASE databases were searched from their respective inceptions in October 2013. **Review methods:** Exercise training studies with at least one outcome measure regarding wheelchair propulsion capacity were included. In this study wheelchair propulsion capacity includes four parameters to reflect functional wheelchair propulsion: cardio-respiratory fitness (aerobic capacity), anaerobic capacity, muscular fitness and mechanical efficiency. Articles were not selected on diagnosis, training type or mode. Studies were divided into four training types: interval, endurance, strength, and mixed training. **Methodological quality** was rated with the PEDro scale, and the level of evidence was determined. **Results:** The 21 included studies represented 249 individuals with spinal-cord injury (50%), various diagnoses like spina bifida (4%), cerebral palsy (2%), traumatic injury, (3%) and able-bodied participants (38%). All interval training studies found a significant improvement of 18-64% in wheelchair propulsion capacity. Three out of five endurance training studies reported significant effectiveness. **Methodological quality** was generally poor and there were only two randomised controlled trials. **Conclusion:** Exercise training programs seem to be effective in improving wheelchair propulsion capacity. However, there is remarkably little research, particularly for individuals who do not have spinal-cord injury.

[PMID: 24615862](#) [PubMed - as supplied by publisher]

9. J Health Psychol. 2014 Mar 6. [Epub ahead of print]

Barriers and facilitators to participation in physical activity: The experiences of a group of South African adolescents with cerebral palsy.

Conchar L1, Bantjes J, Swartz L, Derman W.

Participation in regular physical activity promotes physical health and psychosocial well-being. Interventions are thus needed to promote physical activity, particularly among groups of individuals, such as persons with disability, who are marginalised from physical activity. This study explored the experiences of a group of South African adolescents with cerebral palsy. In-depth semi-structured interviews were conducted with 15 adolescents with cerebral palsy. The results provided insight into a range of factors that promote and hinder participation in physical activity among adolescents with cerebral palsy in resource-scarce environments.

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10. Med Oral Patol Oral Cir Bucal. 2014 Mar 8. [Epub ahead of print]

Consensus Report of the XI Congress of The Spanish Society of Odontology for the Handicapped and Special patients.

Machuca-Portillo G1, Cabrerizo-Merino C, Cutando-Soriano A, Giménez-Prats MJ, Silvestre-Donat FJ, Tomás-Carmona I.

This article summarizes the findings of consensus of the XI congress of the SEOEME) All of these conclusions are

referring to the review articles responsible to the general rapporteurs in order to bringing up to date knowledge with regard to the use of implants in patients medically compromised and with special needs and, in the dental management of autism and cerebral palsy, in the dental treatment of patients with genetic and acquired haematological disorders, the dental implications of cardiovascular disease and hospital dentistry.

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11. Med Oral Patol Oral Cir Bucal. 2014 Mar 8. [Epub ahead of print]

Dental treatment under general anesthesia in a group of patients with cerebral palsy and a group of healthy pediatric patients.

Escanilla-Casal A1, Aznar-Gómez M, Viaño JM, López-Giménez A, Rivera-Baró A.

This is a comparative study between two groups, one of healthy children and the other of children with cerebral palsy, which underwent dental treatment under general anesthesia at Hospital Sant Joan de Déu Barcelona. The purpose of the study was to compare and determine oral pathology, frequency, severity and postoperative complications in pediatric patients with and without an underlying disease which undergo a dental treatment under general anesthesia.

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11. Clin Nutr. 2014 Feb 20. pii: S0261-5614(14)00052-1. doi: 10.1016/j.clnu.2014.02.007. [Epub ahead of print]

Differences in body composition according to functional ability in preschool-aged children with cerebral palsy.

Walker JL1, Bell KL2, Stevenson RD3, Weir KA4, Boyd RN5, Davies PS6.

BACKGROUND & AIMS: Altered body composition is evident in school children with cerebral palsy (CP). Fat free mass and fat mass amounts differ according to functional ability and compared to typically developing children (TDC). The extent to which body composition is altered in preschool-aged children with CP is unknown. We aimed to determine the fat free mass index (FFMI) and body fat percentage (BF%) of preschool-aged children with CP and investigate differences according to functional ability and compared to TDC. **METHODS:** Eighty-five children with CP (68% male) of all functional abilities, motor types and distributions and 16 TDC (63% male) aged 1.4-5.1 years participated in this cross-sectional study. Body composition was determined via isotope dilution. Children with CP were classified into groups based on their Gross Motor Function Classification System (GMFCS) level. Statistical analyses were via ANOVA, ANCOVA, post-hoc Tukey HSD tests, independent t-tests and multiple regressions. **RESULTS:** There were no significant differences in FFMI or BF% when comparing all children with CP to TDC. Children classified as GMFCS levels III, IV and V had significantly lower FFMI levels compared to children classified as GMFCS I and II ($p < 0.05$). Children of GMFCS IV and V had the highest mean (\pm SD) BF% of all children (24.6% (\pm 10.7%)), significantly higher than children of GMFCS I and II (18.6% (\pm 6.8%), $p < 0.05$). **CONCLUSIONS:** Altered body composition is evident in preschool-aged children with CP, with a trend towards lower FFMI levels and greater BF% across functional ability levels from GMFCS I to V. Further research is required to determine optimal body composition parameters and investigate contributing factors.

CLINICAL TRIAL REGISTRY: Australian New Zealand Clinical Trials Registry (ANZCTR) number: ACTRN12611000616976.

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12. J Neuroeng Rehabil. 2014 Mar 8;11(1):34. [Epub ahead of print]**Autonomic responses to correct outcomes and interaction errors during single-switch scanning among children with severe spastic quadriplegic cerebral palsy.**

Leung B, Chau T.

BACKGROUND: The combination of single-switch access technology and scanning is the most promising means of augmentative and alternative communication for many children with severe physical disabilities. However, the physical impairment of the child and the technology's limited ability to interpret the child's intentions often lead to false positives and negatives (corresponding to accidental and missed selections, respectively) occurring at rates that frustrate the user and preclude functional communication. Multiple psychophysiological studies have associated cardiac deceleration and increased phasic electrodermal activity with self-realization of errors among able-bodied individuals. Thus, physiological measurements have potential utility at enhancing single-switch access, provided that such prototypical autonomic responses exist in persons with profound disabilities. **METHODS:** The present case series investigated the autonomic responses of three pediatric single-switch users with severe spastic quadriplegic cerebral palsy, in the context of a single-switch letter matching activity. Each participant exhibited distinct autonomic responses to activity engagement. **RESULTS:** Our analysis confirmed the presence of the autonomic response pattern of cardiac deceleration and increased phasic electrodermal activity following true positives, false positives and false negatives errors, but not subsequent to true negative outcomes. **CONCLUSIONS:** These findings suggest that there may be merit in complementing single-switch input with autonomic measurements to improve augmentative and alternative communications for pediatric access technology users.

[PMID: 24607065](#) [PubMed - as supplied by publisher]

13. Rehabil Psychol. 2014 Mar 10. [Epub ahead of print]**Gender and Nurturance in Families of Children With Neurodevelopmental Conditions.**

Shapiro DN, Dixon-Thomas P, Warschausky S.

Objective: This study tested the hypothesis that gender differences in parent-reported nurturance of children would be attenuated in families of children with neurodevelopmental conditions (NDCs). **Method:** In this cross-sectional study, participants included 49 (29 male) children diagnosed with an NDC and 60 (30 male) typically developing (TD) children. Children in the NDC group had a diagnosis of cerebral palsy (CP; n = 41) or spina bifida (SB; n = 8). Parental nurturance was measured using the nurturance subscale of the Parenting Dimensions Inventory (PDI; Power, 1991). Data were analyzed using a 2 × 2 (gender × diagnosis) analysis of covariance (ANCOVA) with child age as the covariate. **Results:** As a simple main effect, parents reported more nurturing behavior toward TD girls than TD boys. However, girls with an NDC received less nurturance, thereby eliminating the gender difference in parental nurturance in the NDC sample. This pattern was reflected in the larger ANCOVA as a 2-way interaction between diagnosis and gender. Group differences in other PDI subscales were not statistically significant. **Conclusions and Significance:** This pattern of results suggests that the parents of girls with NDCs may be less nurturing toward them, thereby attenuating gender differences observed in families with TD children. Findings highlight the need for more research on the gendered dynamics in families with a child with an NDC to develop systemic models of family functioning and targeted parenting interventions for this group. (PsycINFO Database Record (c) 2014 APA, all rights reserved).

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Prevention and Cure

14. *Dev Med Child Neurol.* 2014 Mar 13. doi: 10.1111/dmcn.12430. [Epub ahead of print]

Risk of cerebral palsy in relation to pregnancy disorders and preterm birth: a national cohort study.

Trønnes H1, Wilcox AJ, Lie RT, Markestad T, Moster D.

AIM: To assess the risk of developing cerebral palsy in relation to pregnancy disorders and preterm birth.

METHOD: By linking the Medical Birth Registry of Norway to other national registries, we identified all live births in Norway from 1967 through to 2001. Risks of cerebral palsy (CP) after preterm delivery and pregnancy disorders were estimated in different gestational age groups. RESULT: In total, 1 764 509 children delivered at 23 to 43 weeks' gestation were included. The prevalence of CP was 1.8 per 1000 births. Absolute risk of CP was 8.5% among children born at 23 to 27 weeks' gestation, 5.6% at 28 to 30 weeks, 2.0% at 31 to 33 weeks, 0.4% at 34 to 36 weeks, and 0.1% thereafter. Placental abruption, chorioamnionitis, prolonged rupture of membranes, intrauterine growth restriction, pre-eclampsia, multiple births, placenta previa, bleeding, cervical conization, and congenital malformation were all associated with CP. Before 32 weeks' gestation, absolute risk of CP was highest with chorioamnionitis (9.1%) and lowest with pre-eclampsia (3.1%). Among those born after 31 weeks, the absolute risk of CP was more consistently (but also more slightly) increased with a recorded pregnancy disorder.

INTERPRETATION: Early delivery and pregnancy disorders were both strong risk factors for CP. The added risks with recorded pregnancy disorders varied within categories of gestational age.

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15. *Brain.* 2014 Mar 10. [Epub ahead of print]

Dopamine transporter deficiency syndrome: phenotypic spectrum from infancy to adulthood.

Ng J1, Zhen J, Meyer E, Erreger K, Li Y, Kakar N, Ahmad J, Thiele H, Kubisch C, Rider NL, Holmes Morton D, Strauss KA, Puffenberger EG, D'Agnano D, Anikster Y, Carducci C, Hyland K, Rotstein M, Leuzzi V, Borck G, Reith ME, Kurian MA.

Dopamine transporter deficiency syndrome due to SLC6A3 mutations is the first inherited dopamine 'transportopathy' to be described, with a classical presentation of early infantile-onset progressive parkinsonism dystonia. In this study we have identified a new cohort of patients with dopamine transporter deficiency syndrome, including, most significantly, atypical presentation later in childhood with a milder disease course. We report the detailed clinical features, molecular genetic findings and in vitro functional investigations undertaken for adult and paediatric cases. Patients presenting with parkinsonism dystonia or a neurotransmitter profile characteristic of dopamine transporter deficiency syndrome were recruited for study. SLC6A3 mutational analysis was undertaken in all patients. The functional consequences of missense variants on the dopamine transporter were evaluated by determining the effect of mutant dopamine transporter on dopamine uptake, protein expression and amphetamine-mediated dopamine efflux using an in vitro cellular heterologous expression system. We identified eight new patients from five unrelated families with dopamine transporter deficiency syndrome. The median age at diagnosis was 13 years (range 1.5-34 years). Most significantly, the case series included three adolescent males with atypical dopamine transporter deficiency syndrome of juvenile onset (outside infancy) and progressive parkinsonism dystonia. The other five patients in the cohort presented with classical infantile-onset parkinsonism dystonia, with one surviving into adulthood (currently aged 34 years) and labelled as having 'juvenile parkinsonism'. All eight patients harboured homozygous or compound heterozygous mutations in SLC6A3, of which the majority are previously unreported variants. In vitro studies of mutant dopamine transporter demonstrated multifaceted loss of dopamine transporter function. Impaired dopamine uptake was universally present, and more severely impacted in dopamine transporter mutants causing infantile-onset rather than juvenile-onset disease. Dopamine transporter mutants also showed diminished dopamine binding affinity, reduced cell surface transporter, loss of post-translational dopamine transporter glycosylation and failure of amphetamine-mediated dopamine efflux. Our data series expands the clinical phenotypic continuum of dopamine transporter deficiency syndrome and indicates that there is a phenotypic spectrum from infancy (early onset, rapidly progressive disease) to childhood/adolescence

and adulthood (later onset, slower disease progression). Genotype-phenotype analysis in this cohort suggests that higher residual dopamine transporter activity is likely to contribute to postponing disease presentation in these later-onset adult cases. Dopamine transporter deficiency syndrome remains under-recognized and our data highlights that dopamine transporter deficiency syndrome should be considered as a differential diagnosis for both infantile- and juvenile-onset movement disorders, including cerebral palsy and juvenile parkinsonism.

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16. J Pak Med Assoc. 2014 Jan;64(1):103-7.

Risk factors and types of cerebral palsy.

Bangash AS1, Hanafi MZ2, Idrees R2, Zehra N3.

OBJECTIVE:To determine the risk factors and associated types of cerebral palsy in a squatter settlement of Karachi. **METHODS:** The pilot cross-sectional study was conducted in Karachi during 2010 and 2011. Data was collected through an interviewed questionnaire from the mothers of cerebral palsy victims children from a population of 6000. Sample sizes of 20 pre-diagnosed victims were selected through snowball sampling. SPSS 20 was used for statistical significance. **RESULTS:** The mean age of the 20 children was 8.7 +/- 6.4 years. Of them, 16 (80%) were males and 4 (20%) were females. Major risk factors identified were; home and assisted delivery 5 (75%), consanguinity 10 (50%), infections 8 (40%) and lack of antenatal care 6 (30%). Out of 20 cases, 15 (75%) had spastic type of cerebral palsy, which was further classified as diplegia 7 (35%), quadriplegia 6 (30%) and hemiplegia 2 (10%). Mixed and dystonic types were found in 3 (15%) and 2 (10%) children respectively. **CONCLUSION:** Important risk factors identified were home delivery, consanguinity and infections during pregnancy. Spastic type of cerebral palsy was the most common type in the study population.

[PMID: 24605730](#) [PubMed - in process]