

Hand function in children with cerebral palsy undergoing Intensive neurophysiological rehabilitation.

Study protocol

May 15, 2017

Translated into English from Ukrainian to English by principal investigator Oleh Kachmar

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Introduction

Cerebral palsy (CP) is the most common cause of childhood physical disability with an estimated prevalence of 2.11 per 1000 live births.¹ Limited hand function is a common disabling impairment in children with CP and a strong predictor of a child's restricted ability to participate in daily activities.² Upper limb disorders in CP include weakness, sensory impairment, spasticity, and dystonia.³ Children with CP experience difficulties with grasping and releasing objects due to an imbalance between wrist flexors and extensors.⁴ Everyday activities, self-care, and domestic tasks are often hard for them to accomplish.

Growing evidence suggests that high-intensity therapies for upper limb dysfunction in children with CP are more effective compared to usual, less intensive care, suggesting that the dosage, i.e., treatment amount and frequency, is as important as the treatment itself.^{5,6,7,8} The most promising options are constraint-induced movement therapy (CIMT)—a unimanual training—and hand-arm bimanual intensive therapy (HABIT)—a bimanual training.^{9,10} Both therapies improve unimanual movements, bimanual capacity, and movement efficiency in children with CP.^{11,12} Several studies suggested that structured bimanual training might drive neuroplasticity by increasing the size of the motor map of the affected hand and improving action planning of the upper limb movements.^{13,14}

Setting rehabilitation goals that are realistic and relevant to the daily life is also essential.¹⁵ Intensive, activity-based, goal-directed functional interventions were more effective than standard care in improving hand function, self-care, and individualized outcomes.^{11,16} An intensive program combining group and individual sessions resulted in a high rate of goal attainment, even though the participants were of different age and functional levels and had different goals.¹⁷ Studies also showed that a short high-intensity course combining interventions for upper and lower extremity improved function of both upper and lower extremity in children with unilateral and bilateral spastic CP more effectively than conventional treatment (delivered over a longer period).^{18,19}

The interventions that proved to be effective share similar features—they are high-dose, repetitive, incrementally challenging, goal-oriented, and functional. So, the attention of researchers and clinicians is currently focused on the exploration of intensive, goal- and function-oriented approaches addressing multiple limitations.

One of such approaches is Intensive Neurophysiological Rehabilitation System (INRS)—a two-week multicomponent course that combines several interventions for both upper and lower limbs and addresses different functional goals.²⁰ The interventions include age-appropriate unimanual and bimanual fine and gross motor activities, the difficulty of which gradually increases. INRS components are as follows: physical therapy, occupational therapy, spinal manipulative therapy, gait training, joint mobilization, strength training, full body massage, computer game therapy, suit therapy, and group sessions of rhythmic gymnastics. The course is flexible in that regard that the dose of each component and the components themselves are modified to suit a patient's condition and goals. It is intensive but child-friendly at the same time, because the activities involve elements of play. Several studies reported positive effects of INRS on children with CP, including improvement of gross motor functions, reduction of spasticity, increase in passive range of motions, and development of fine motor skills.^{21,22}

This study was prompted by promising results from studies on different types of intensive goal-oriented training for upper limb dysfunction and positive effects of intensive INRS use in patients with CP. The aim of present study was to assess changes in hand function in children with bilateral spastic forms of cerebral palsy after the two-week treatment with INRS.

Materials and methods

A prospective case series study design would be used. Conducting the study and inclusion of children with disabilities will be approved by the local Ethical Commission. Participants and their legal representatives will receive comprehensive information about the intervention and study

design. Written informed consent would be obtained from legal representatives. Verbal assent will be given by participants, when appropriate, based on age and cognitive skills.

Participants

Patients of the tertiary care facility will be assessed for eligibility. Inclusion criteria were spastic forms of CP, bilateral impairment, age 6 to 15 years, Manual Ability Classification System (MACS) level I-III. Exclusion criteria are moderate to severe cognitive deficit, uncooperative behavior, severe contractures of upper extremity joints, previous hand surgery, and epilepsy with frequent seizures. After applying the exclusion criteria, 32 patients (mean age 10.2 years; 17 males) were selected for the study. The dominant hand was established based on parental report and medical history.

Intervention

All participants will undergo a two-week Intensive Neurophysiological Rehabilitation System (INRS) course in the tertiary care facility. INRS is a novel intensive approach that combines several multi-faceted rehabilitative components to improve functioning and quality of life of children with CP.²⁰ INRS involves active and passive treatment components that complement and reinforce each other. Active components are aimed at acquisition of new skills and abilities while passive components prepare physiological background for skills development.

INRS program course is tailored individually based on age, motor abilities, and functional goals within International Classification of Functioning, Disability and Health (ICF) domains. The program course includes repeated procedures in which the difficulty of required movements is gradually increased. Repetitive tasks with gradually increasing complexity and motivation reportedly thought to stimulate plastic changes in the brain.²⁴ The components are developed to be child-friendly and take into account the motivational aspect of rehabilitation. They focus on the functional performance of daily life activities along with improvement of the postural control and locomotion.

The course involves 11 days of treatment (6 days during the first week, and 5—during the second), 4 to 5 hours daily and up to 55 hours in total. Treatment is provided by a multidisciplinary team of experienced medical doctors, physical therapists, and nurses. The quality of every component delivery is controlled by the supervisor. No adverse events were reported during the intervention.

Outcome Measures

The same evaluators will assess each child two times: before and after the two-week course of INRS. The evaluators would not be involved in treatment.

Primary outcome measure

The Jebsen-Taylor hand function test (JTHFT) assesses performance of seven manual tasks mimicking common activities of daily living.²⁵ The JTHFT reflects changes in the category Dexterity of the Body Functions and Body Structures domain of the ICF, and the subcategory Carrying, moving or manipulating objects of the Activity domain of the ICF.²⁶ Although the JTHFT was primarily designed for adult patients, it was also proved a reliable and responsive tool for tracking short-term changes in children with CP.^{27,28}

The test consists of seven subtests: writing a simple sentence, card turning, picking up small common objects, spooning five kidney beans into a bowl, stacking four checkers, moving large light objects, and moving large weighted objects. The writing task was excluded because some children were too young.²⁷ The supposedly weaker hand, usually the more affected, non-dominant hand was evaluated first.

JTHFT measures the time it takes the child to complete the standardized functional tasks with each hand and better performance. Maximum two minutes are allowed for a subtest. The total score for a hand is a sum of all subtest times for that hand. A lower score indicates better performance.

Secondary outcome measures

ABILHAND-Kids questionnaire is a measure of manual ability for children with upper limb impairments.²⁹ Manual ability refers to the Activity domain of the ICF. A parent or a caregiver is asked to assess how difficult it is for a child to accomplish a set of 21 selected activities of daily living on a three-level scale (impossible, difficult, easy). When giving the assessment, the responder should focus on the result of the activity and not on the method used to accomplish it (e.g. it is not important if the child uses one or both hands).

The items of the questionnaire are presented randomly to avoid systematic effect. The questionnaire was designed using the Rasch measurement model that allows converting raw scores into linear units on a unidimensional scale. We used the Ukrainian version of ABILHAND-Kids that had been adapted and validated for the Ukrainian population.³⁰

Manual dexterity was evaluated with **Box and Blocks test (BBT)**, a valid and reliable measure for different categories of patients with neurological impairments ^{28,31}. For children with cerebral palsy, the test was proved to be sensitive in short-term interventions ²⁸. BBT covers Dexterity category of the Body Functions and Body Structures domain of the ICF. The child has one minute to carry as many blocks from one compartment of the box to another as possible ³¹. The scores of BBT are the number of blocks, carried over by each hand.

Grip strength was measured with a Jamar hand dynamometer (Performance *Health*, USA). This device has good test-retest and interrater reliability according to a systematic review for upper extremity strength measurement in children with CP ³². Hand-grip dynamometry encompasses the Muscle strength functions category of the Body Functions and Body Structures domain of ICF. Three measurements of maximum voluntary contractions were taken for each hand, and the best result was recorded.

Statistical Analysis Plan

IBM SPSS v23 software will be used to perform statistical analysis. Normality will be assessed using the Kolmogorov-Smirnov test.

For normally distributed variables descriptive statistics includes means with standard deviation (SD) values. Baseline and post-intervention results will be compared using the Paired Sample two-tailed T-Test with 95% confidence interval. In calculations, statistical significance level (α) was taken as 5% and values with $p < 0.05$ expressed statistical significance.

In case of non-normal distribution, nonparametric statistics would be used. Medians with interquartile range will be used for descriptive statistics, Wilcoxon signed-rank test would be used to compare baseline and post-intervention data

Informed Consent Form



Informed consent for parents of children participating in the study

We ask for your participation in the research project “Hand function in children with cerebral palsy undergoing Intensive neurophysiological rehabilitation.” which aims to assess the effect of Intensive Neurophysiological Rehabilitation system on arm function in patients with cerebral palsy.

Our previous studies describe an increase in arm strength and dexterity in a group of children with cerebral palsy after a course of Intensive Neurophysiological Rehabilitation. But other important components of hand function (such as hand involvement in a child's daily activities) were not evaluated in this study.

Procedure

Participating children will be additionally assessed two times: before and after the two-week treatment course in the International Clinic of Rehabilitation. The evaluators would not be involved in treatment.

The evaluation will consist of four components:

- The Jebsen-Taylor test is a tool for assessing a child's performance of a list of tasks that are often used in everyday life.
- Abilhand-Kids questionnaire is a tool for assessing the child's ability to perform daily activities that require the participation of the upper extremities.
- Test "Box and Blocks" - assessment of dexterity.
- Dynamometry - assessment of hand strength using a Jamar wrist dynamometer.

No component of the examination causes pain or other discomfort to the patient.

Participation in the study is completely voluntary, there is no risk of participation in the study. Participants or their representatives have the right to withdraw at any time without explanation.

The research team guarantees each patient's confidentiality and guarantees that the research's results will not be used for other purposes than those detailed in the project.

Personal data will not be collected, stored or distributed. All personal information will be anonymous and encrypted. In any case, neither the name nor other personal data will be disclosed in research articles or at the request of third parties.

Certificate of consent

I have been informed, in a comprehensive way, about these and other aspects of my possible participation in the project (through the information sheet) and I have had the opportunity to ask questions and to resolve any doubt that I might have. I have decided to participate in the project knowing that this decision does not affect the care and treatment that I am receiving in the Clinic, and I am also aware that I can withdraw from the study at any time.

Also, my son/daughter/dependent has been informed and has understood the procedures of the study. All his/her questions and doubts have been answered and he/she has voluntarily consented to participate in the project.

I have been informed that my personal data will be properly stored and processed.

I freely and voluntarily agree that my son/daughter/dependent participates in the project. And that is why I explicitly authorize this consent form.

Name of the participant: _____

Name of parent or legal guardian: _____

Parent or legal guardian's signature: _____

Date: " ____ " _____ 20

Statement of the contact person: I have explained the nature and purpose of this research, as well as the possible risks to participants. I have answered the questions made by the interviewee. I will provide the parents or guardians with a copy of the informed consent.

Contact person name and signature _____