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Development, validation and reliability testing of a sensory outcome measure for hypoxia induced cerebral palsy children: A study Protocol

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Abstract

Background: The motor disorders of cerebral palsy suffering from hypoxic ischemic encephalopathy are associated with sensory perception impairment along with other impairment like cognition, communication, aberrant behavior, brain stems, or a combination of these features. There are several scales available to evaluate muscle tone, balance, gait, motor power, gross motor function and intelligence but no scale is available till date to measure the sensory issues in children of HIE for Indian population.

Objectives:

The objectives of the study are to measure the sensory issues in children of HIE and to validate the scale for its content, reliability and minimum detectable change (MDC).

Methods:

In this study there are four phases: First phase is concerned with the development of scale item and its validation. Second phase is to check the reliability of the scale (test re-test reliability and inter rater reliability). In third phase we will estimate standard error of measurement (SEM) and in last fourth phase minimal detectable change (MDC) at 95% Confidence Interval (CI) will be checked.

Data analysis:

All the items in scale will be check for reliability and criterion-related validity test. Minimum 60 children with HIE will be evaluated with the scale for two times within two weeks to establish test-retest reliability. Cronbach's alpha will be use for internal consistency and Intra class correlation coefficient for test retest reliability for determination of the degree of consistency of items present in the scale.

Conclusion:

The scale, thus formed will be the appropriate outcome measure for documenting patient outcome in India.

Keywords: hypoxic ischemic encephalopathy, cerebral palsy, scale formulation, sensory outcome.

Introduction

Hypoxic ischemic encephalopathy (HIE) is a severe birth complication mainly affecting the neonates. It occurs due to brain damage by improper blood supply to brain causing brain asphyxia during child birth, before child birth and after child birth which further causes CP. Several studies suggest that more than half of the sufferers with HIE will expire by the age of 2 years, and those who are live will suffer with many problems like intellectual retardation, seizures, and CP.[1] HIE is a set of condition that causes permanent disturbance of the development of posture as well as limitations in performance. These attributes are non-progressive which take place in the developing brain of a fetus or infant. Some sensory disturbances like sensitivity, memory, statement, conduct, epilepsy, and secondary musculoskeletal problems are also seen along with motor disturbances.[2]

Population based studies suggest that the birth occurrence of CP is about 2 to 9 per 1,000 live births. [3] HIE is a common pediatric disorder.[1] It is a chronic condition which is non progressive in nature (static). Children suffering from HIE faced so many problem such as epilepsy, difficulties in feeding, visual problems and hearing impairment.[4] Due to so many problems like abnormal change in muscle tone, posture there is limitation in activity. Although CP is unprogressive disease but medical features may progress as the brain develops. [3]

The exact cause of HIE not recognized but several past history comprise cord prolaps, uterine burst, abruptio placenta, placenta previa, maternal hypotension, breech appearance, or shoulder dystonia. The symptoms of HIE which are seen in neonates are: fetal arrhythmia, poor umbilical cord gases (pH < 7.0 or base deficit ≥ 12 mmol/L), low Apgar scores, presence of meconium stained fluid, necessity for respiratory help just after delivery of child.. The pathological changes occurs in HIE are due to abnormal blood and oxygen supply to the cerebral part of brain.[5]

Factors causing HIE are intrauterine factor like restriction in fetal growth, placental vascular disorders, Some peripartum factors like placental abruption and some events at the neonatal period like intraventricular hemorrhage and neonatal stroke.[3] Ischemia causes excessive production of cytokines, oxygen free radical, deficiency of maternal growth factor, extracellular matrix alteration and increased production of glutamate, triggers the excitotoxic cascade.[6]

So many neurodegenerative conditions, including metabolic and inherent disorders may shows similar sign and symptoms like CP for example anterior horn cell disorder, peripheral neuropathy, defect in neuromuscular transmission, Duchene muscular dystrophy (DMD), Dopa responsive dystonia(DRD), Meta chromatic leucodystrophy, Adreno leucodystrophy, adrenomyelo neuropathy, heterogeneous degenerative disorder of the cervical cord and Lesc Nyha Ataxia telangiectasia (AT), X linked spinocerebellar ataxia, Pontocerebellar atrophies / hypoplasias, carbohydrate deficient glycoprotein (CDG), Worster Drought syndrome.[7] Clinical features of CP/HIE comprise some unconstructive phenomenon such as weakness of skeletal muscles and developmental mile stones are

delayed for several months and some positive phenomenon like velocity dependent increased muscle tone with spasticity, Clonus, rigidity, spasms and hyper reflexia.

A smaller percentage of children with CP /HIE may exhibit some other symptoms as abnormal movements like athetosis, chorea, and dystonia. Mixed CP are those who shows a combination of features such as epilepsy, feeding, nutrition, growth problem, mental retardation, bladder dysfunctions, bowel dysfunction, drooling, sleep disturbances, hearing loss, visual abnormalities, orthopedic associated sensory impairment in CP are poorly understood.[8] Along with the motor disturbance some other symptoms like difficulty in understanding, language problem , sensory awareness, problem in behavior, fits, or a group of these features can be seen with CP child.[9]

CP can be classified on the basis of Pathophysiology as if the cortical involvement is there choreoathetosis occurs, cerebellar involvement resulting in abnormal movements, if involvement of basal ganglial occurs hypotonia exhibits.[4] CP can also be classified according to the type of the motor disturbance like Paresis, spastic, Hypotonic, Diatonic, Dyskinetic or ataxic type. It can also be further classified according to the parts of the body affected as paraplegics, hemiplegics or quadriplegics.[10] The sign and symptoms of the Pyramidal lesions are increase in tone of skeletal muscles, deep-tendon reflexes and plantar response is extensor. Lesions in extra pyramidal system show abnormal involuntary movements along with difficulty in coordination and maintaining the posture. [9]

The diagnosis of CP is based on laboratory investigations along with some sign and symptoms. The symptoms like developmental delay, walking on toe, cortical thumb, smaller head size, fits, bad temper, and difficulty in sucking can be an early indicator. Evaluation of child suggests scissoring of the lower limb and retained primitive reflexes. More precious investigations involved CT and MRI of brain. In an around 63% of cases Brain CT may be abnormal. [4]

Assessments have to be done by medical information which include a comprehensive history along with interview of parents or care taker, examination of nervous system as well examination of motor system. Ambulatory status and motor functioning (rolls over, independent sitting, sliding, supported standing, independent standing, assistance walking, walks with assistive devices, independent walking) have to be assessed on clinical examination. Sensory testing included assessment of superficial and deep sensations.[11] Management includes medicinal treatment as antispasmodics to relax tight muscles, anti cholinergic medication for managing uncontrollable body movements and anticonvulsants for the treatment of seizures. Sometimes orthopedic and neural surgeries are also required for joints, muscles, tendons and nerves. Casts, splints & braces may also be required for assistance.[12] There are so many scales are available for assessment of gross motor functioning, mobility, balance, disability in physiotherapy.

Sensory processing is a combined effort defined as sensory information by nervous system; it includes the function of the peripheral nervous system, central nervous system and information about sense organs. Brain receives sensory impulse from the surroundings and also from the body itself. Brain utilizes this information to recognize experiences and organize them for the suitable response. Persons are able to respond automatically, competently, and at ease to the specific sensory stimuli by this process. Motor responses and sensory inputs are interrelated. The capability to arrange

information from the sensory system to make a modifying reaction is known as sensory integration. Sensory abnormalities affect the motor functioning of an individual. [13]

Rationale of the Study

The present scales could not give all-inclusive evaluation for sensory issues in HIE children for Indian population. That's why, there is a requirement to develop a scale to measure the all domain in HIE children. The purpose of this present study is to develop a sensory outcome measure for assessment of sensory issues in HIE children using a systematic approach.

Objectives: The objectives of the study are:

To develop an outcome measure of sensory testing in children with Hypoxia induced cerebral palsy children.

To evaluate the validation of the Sensory outcome measure for Hypoxia induced cerebral palsy children.

To evaluate reliability of Sensory outcome measure for Hypoxia induced cerebral palsy children

Methods

Trial Design

The study would be a cross-sectional study for the validation of scale and reliability testing. The study protocol is approved by Institutional Ethics Committee of Swami Vivekanand Subharti University, Meerut, Uttar Pradesh, India.

Four Steps of Construct Development

STEP 1: Construct Definition

STEP2: Item Generation and Validation

- **Extensive literature review and focus group**
- **Grouping of items**
- **Content validation (by experts panel)**
- **Rough draft**
- **Pilot study**
- **Revision**
- **Final format**

STEP 3: Testing of Reliability

INTERNAL CONSISTENSY BY CRONBACH,S ALPHA	**TEST RE TEST** • SESSION 1 (By principal investigator) • 7 DAY	**INTRARATER** • SESSION 1(By principal investigator) • BREAK OF	**INTERRATER** • SESSION 1 RATER 1 RATER 2 • 7 DAYS

	BREAK INTERVAL • SESSION 2 (Again by principal investigator)	SHORT PERIOD • SESSION 2(Again by principal investigator)	BREAK INTERVAL • SESSION 2 RATER 1 RATER 2 • MEAN VALUE

STEP 4: MDC

- **SEM**
- **MDC**

Material used: Sensory tool kit, CP chair (for severely physically handicap)

Participants

The target population is children suffering from hypoxia induced cerebral palsy. Participating children of 5-13 years of age group are selected for the trial.

Recruitment

Children are recruited from community advertisement, news paper ads and through Pediatric department of Chhatrapati Shivaji Swami Vivekanand Hospital, Meerut, Uttar Pradesh, India. A Poster explaining the research project is being displayed at pediatric clinics. Interested participants are invited to call by their parents and care giver.

Determination of eligibility is achieved by screening done by pediatrician at pediatric department of research site.

S. No.	Inclusion criteria	Exclusion criteria
1.	Children suffering from HIE.	Children who having some associated conditions (congenital malformations, cardiac pathology)
2.	Age ranges from 5-13 years.	Etiological factor other than HIE.
3.	Both male and female	Hearing impairment, Mental retardation, Visually impaired.

Sampling method: Purposive

Research Site

The study is conducted at research center of Subharti College of Physiotherapy, Swami Vivekanand Subharti University Meerut, Uttar Pradesh, India

Procedure

Step 1: Construct definition: Defining about the construct

Step 2: Item generation: Item development step will consists of seven sub-steps. These are item generating, grouping items (designing the scale), content validation, rough draft, pilot study, revision and final format. This step will provide all items related to sensory issues in HIE patients using the interview of the care giver and according the literature available.

Literature search

The literature for English language will be searched in Pub Med, Cochrane Library and goggle scholar databases in between January 1990 and February 2020. The proposed key words which are to be used are hypoxic ischemic encephalopathy, sensory scale, proprioception, kinesthesia, vibration, two point discrimination and sensory assessment scale.

For direct interview, twenty parents of HIE children will be approached at home or OPD by the investigator and data was collected. These parents will be requested to produce the items that are significant to assess the sensory issues. First, the parents will be requested to state different items related to sensory problems which they consider in their children. Motivate the parents to answer highest number of items which they feel correct on the basis of their experience. Second, the parents will be given a set of pool of items searched from the literature and will be requested to add more number of items which was not given in the literature. By this we will be chosen the highest number of items, which the parents felt that need to be in the scale. After generation of items from the literature and interviews, the items will be pooled together and duplicacy has to be removed.

Grouping item

After generation of items by literature search and parent interviews, the items will be grouped under the domains.

Content validation

The grouping of items would be followed for its content validation by the panel of experts (sample of 10). Agreements between experts have to evaluate for the content validity of items. The criteria which to be include the items and domains will be fixed at 80% i.e., the items will be included only if eight or more experts give positive response.

Rough draft

The items will be accepted, modified or removed based on the level of agreement between experts. The scoring criteria for the scale will be formed to represent the sensory issues on a 4-point scale ranging from 0 to 3, where 0 is no impairment and 3 is severe impairment. The middle scores represent 1 (Mild impairment), 2 (Moderate impairment). The scoring options will be designed according to the items under each domain. The total score of the scale will be 100% which means extremely severe impairment of sensory issues in HIE children.

Pilot study

Pilot will be done to assess the comprehensibility, acceptability, complexity, administrative difficulties, and frequency of responses and sequence of item arrangements in the scale.

Revision

It will be decided to eliminate the item, if the frequency of response is less than 30% for the item. The modification of items and scoring criteria will be made after discussion with experts involved in the content validation. After that consensus will be obtained regarding the domains, items and its arrangement in the scale, the final format will be formed. This scale with modified items and domains will be subjected to the next step of validation.

Final draft

The final draft of the proposed will be made after incorporating the above changes. The suitable name will be fixed to the scale.

Phase 2: Reliability testing

Check for internal consistency

The sample size will be calculated from the pilot study data. The new scale drafted will be administered on the HIEC by investigator. The tester will provide adequate explanation and all the items in the scale have to be scored. The scores will be added. These scores will be used to analyze the internal consistency of items in the drafted new scale using Cronbach's alpha.

Test-retest reliability

Test- retest reliability will be estimated by administrating the scale in HIE children on two periods with minimum duration of seven days and not more than fourteen days by the principal tester.

Intra rater reliability

Intra rater reliability will be estimated by administrating the scale in HIE children on two periods with short intervals by the principal investigator when carryover or practice effects may not be an issue.

Inter rater reliability

Inter rater reliability **w**ill be estimated by two rater in first session and again after an interval with these two raters.

Estimating SEM and MDC at 95% Confidence Interval (CI)

SEM and MDC will be calculated using the formula, SEM = SD x $\sqrt{}$ (1-reliability) and MDC95 = $\sqrt{2}$ x (1.9) x (SEM). Level of Significant will be set at $p \leq 0.05$ to minimize the type-I error. For MCID, responsiveness of the scale will be determined on two occasions after regular conventional physiotherapy treatment of two week duration.

Data analysis

SPSS software IBM Statics 20 version will be used for statistical analysis.

Validity: Delphi method of validation

The rough draft of questionnaire will be send to the panel of 10 experts through email. After receiving the responses content validity ratio will be checked by marking done by experts on three point likert scale related to necessity of items. Content validity index along with scale level content validity of items will be checked by marking done by experts on four point likert scale for relevancy. Clarity of items will also assist with the help of four point likert scale of clarity.

Reliability testing

Internal consistency of items in the drafted new scale will be estimated using Cronbach's alpha (α). Cronbach's alpha ≥ 0.8 will be considered as evidence of acceptable internal consistency. Test retest reliability will be evaluated using Intra class Correlation Coefficient (ICC). ICC will be computed for evaluating the test retest reliability for each domain scores and the total scores. For the purpose of this study, ICC ≥ 0.75 will be considered as evidence of acceptable test–retest reliability. The level of significance will be set at $p \leq 0.05$ for all the statistical analysis. Bland- Altman graph will be used to determine the level of agreement the between sessions.

SEM and MDC at 95% CI

MDC will be calculated using the formulae, SEM = SD x √ (1-reliability) and MDC95 = √2 x (1.9) x (SEM). Test-retest reliability score will be used in place of reliability. Significant level will be set at $p \leq 0.05$ to minimize the type-I error.

Discussion

Infants must be examine for the sensory issues and sensory interventions should be given to them for improvement.[14] It is important to examine the sensory processing challenges in children because the sensory issues may affect the behavior and learning of child.[15] There is weak somatosensory cortical oscillations exhibits in the children with CP.[16] Most of the children with CP exhibit poor tactile perception.[17] Mild tactile deficit in dominant hand and non dominant shows greater sensory impairment.[18] Motor assessment along with the examination of primitive reflexes and postural reactions are essential for early screening.[19] The sensory profile analyzing children's sensory processing patterns.[20] It is helpful for the evaluation of sensory processing abilities in CP and typical children.[21] Sensory processing measure is reliable and valid tool, it can differentiate child with and without sensory processing issues.[22] There is a significant correlation is present between sensory profile and sensory processing measure and have convergent validity of the moderate level for the sensory assessment for the school aged children of Newzeland.[23] For sensory processing distinctiveness in children 15 tests are available to evaluate sensory issues but none of them is for Indian population.[24]

Conclusion

The scale, thus formed will be the appropriate outcome measure for documenting sensory issues in CP children. This Scale will be the first scale which measure the sensory issues in HIE patients. This single scale will provide the overall status of sensory issues in HIE patients. The floor and ceiling effects also have to be determined with the total score. This scale will help in presenting the changes in score of domains before and after treatment. Thus this scale may consider as the best outcome measure for documenting sensory issue in HIEC.

Ethical Statement

The protocol ethical approval will be obtained from Institutional Ethical Committee of Swami Vivekanand Subharti University, Meerut, Uttar Pradesh, India

Conflict of interest

There is no conflict of interest between the authors.

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