Assessment and management of visual perceptual problems in children with cerebral palsy in Cross River State, Nigeria

Roseline Ekanem Duke

Thesis submitted in accordance with the requirements for the degree of Doctor of Philosophy of the University of London SEPTEMBER 2021

Department of Clinical Research

Faculty of Infectious and Tropical Diseases

LONDON SCHOOL OF HYGIENE & TROPICAL MEDICINE

Funded by The Queen Elizabeth Diamond Jubilee Trust/

Commonwealth Eye Health Consortium grant number [LSHTM ITCRZ6814]

I, Roseline Ekanem Duke, confirm that the work presented within this thesis is my own. Where information has been derived from other sources, I confirm this has been indicated in the thesis.

TABLE OF CONTENTS

Table of Figures

List of Tables

Abstract

Cerebral palsy (CP) is the most common physical cause of disability worldwide. Vision impairmentis common in CP and is usually directly associated with the same brain injury which causes the motor problems, the 2 pathways being closely linked anatomically. This is called cerebral visual impairment and can involve 'basic' vision such as visual acuity or visual field or 'higher' visual perception or cognitive vision such as the ability to see moving targets, to pick out a target of interest from a complex scene, visual control of body movement and object recognition. These cognitive visual problems may be termed perceptual visual dysfunction (PVD) and may be missed by conventional ophthalmological assessments. However, the use of inventories of clinical history questions relating to practical aspects of visual perception in everyday life may helping detecting PVD. One example is the Insight Questions Inventory (IQI) in the assessment of PVD which has been studied in the UK, India and Bangladesh. The scores from the IQI inventory for identifying children with PVD have shown some correlation with objective tests of visual perception and with quality of life. Linking responses to the inventory with specifically tailored visual support strategies(IQI VSS), aimed at modifying environment/behaviour to compensate for, or minimize the impact of, the PVD has been piloted in the UK with encouraging results. The IQI has not been used in an African population, nor has it been tested in comparison to more open ended history taking in any population or standard care of observation. In addition, the effectiveness of the linked visual support strategies in impacting quality of life has not been tested through a randomized control trialinvestigation (RCT).

This study was conducted in two phases (phase 1 and 2) between 2016 and 2018 in Cross River state in Nigeria, a lower middle income country. Cross River State is in the South South region of Nigeria. The research was conducted between a collaboration with the Calabar Childrens Eye Centre, Department of Ophthalmology, University of Calabar Teaching Hospital Calabar and the

International Centre for Eye health (ICEH), in London. The researcher is the chief consultant pediatric Ophthalmologist and works with a multidisciplinary team. The Chief investigator from the ICEH group was Richard Bowman and Kathryn Burton as co supervisor, including a team of academic collaborators.

Phase 1, involved the planning of the clinical trial by producing the trial protocol (chapter 4) and the linguistic and cultural modification of the Insight questions inventory (chapter 5). Phase 2, wasa cross sectional study of children with cerebral palsy (with detailed profiling (chapter 6) and assessment of associated co-morbidity (chapter 7) including evaluation of vision (chapter 8) and a randomized controlled study with two arms to investigate whether there is an improvement in quality of life associated with suggesting and implementing visual support strategies for children with CP who have PVD, using IQI (chapter 9).

The primary outcome of the trial assessed after 6 weeks was quality of life using the PedsQL 4.0 generic and PedsQL 3.0 CP modules. Secondary outcomes included subsections of the quality of life scores of the PedsQL 3.0 CP and IQI scores.

Together, these studies indicate that the prevalence of CP in our population is reasonably high (2.2/1000 live births) and that CP is a multimorbid condition, including visual problems which is under-recognized by parents. The presence of co-morbidities was associated with a lower chance ofschool attendance.

We were not able to demonstrate a significant overall improvement in quality of life through the useof IQI VSS. Further work on effective management of co-morbidities associated with cerebral palsyincluding vision which might help their access to education and quality of life are needed. (Chapter10).

List of publications

1. Duke R, Eyong K, Burton K, MacLeoad D,Dutton GN, Gilbert C, Bowman R. The effect of visual support strategies on the quality of life of children with cerebral palsy and cerebral visualimpairment/ perceptual visual dysfunction in Nigeria: study protocol for a randomized controlled trial. Trials. 2019;20:417.

2. Duke R,Torty C,Nwachukwu K, Ameh S, Kim N, Eneli N, Onyedikachi A, Aghaji A, Burton K,Dyet L, Bowman R. Clinical features and aetiology of cerebral palsy in children from Cross River State, Nigeria. Arch Dis Child.2020;0:1-6

3. Duke R, Torty C, Okorie C, Kim M J, Eneli N, Edadi U, Burton K, Tann C, Bowman R.Pattern of comorbidities in school-aged children with cerebral palsy in Cross River State,Nigeria. BMC Pediatrics.2021;21:165

4. Duke RE, Nwachukuw K, Torty C,Okorie U , Kim M, Burton K , Gilbert C , Bowman R. Visual impairment and perceptual visual disorders in children with cerebral palsy in Nigeria. Br J Ophthalmol 2020; 0:1–8.

5. 5. Duke R, Chimaeze T, Kim M, Ameh S, Burton K, Bowman. The effect of Insight Questions Inventory and Visual Support Strategies on carer-reported quality of life for children with cerebral palsy and perceptual visual dysfunction in Nigeria: a randomized controlled trial. Frontiers in human neuroscience. DOI: 10.3389/fnhum.2021.706550

Background/Introduction

2.1 Epidemiology of CP

Children the world over are recognized as a vulnerable group. In November 1989, the convention ofthe right of the child was adopted and opened for signature, ratification and agreement by the United Nations General Assembly resolution. It entered into force on the 2nd of September 1990. Included in Article 29 of the resolution, is the development of the child's personality, talents, mentaland physical abilities to their fullest potential.(1) In the African society, the structure of the community is largely arranged around the lives of its children where traditionally they are viewed as crucial to family survival.(2) The value of the investment in children can be described in emotional, physical, and economic terms. These investments, enable children to do well, contribute to society and have the best quality of life possible. Consequently, this may influence the growth and prosperity of a country.(3) In every society, children are faced with health challenges. The extent to which diseases and ill health affect children depends on the quality of preventive measuresand the use of appropriate and adequate interventions which are influenced by the economic and developmental state of health in which the country is at a particular point in time.(4)

Families respond to acute or chronic ill health, in various ways.(5) Conditions that affect the physical appearance of a child can affect the bond between parents/caregivers and the child and thepride of the parents in the child and cause sorrow.(6-8) Until interventions can occur that improve the quality of the life and the value of the child in the family setting, and larger community, there iseconomic loss and instability in the family and community.(9)

2.1.2 Classification of disease and function

Health can be described in various ways and by the use of various models and concepts.

Physiciansuse the International Classification of Disease (ICD,)(10) to label and identify a disorder, including

the grading of vision, whereas, rehabilitation practitioners explore the functional ability of a personwith a health condition using the International Classification of Functioning, Disability and Health(ICF).(11) The International Classification of Functioning, Disability and Health-Children and Youth (ICF-CY), (12) described by the world health organization, is a framework for describing and organizing information on functioning and disability. Through it, a standard language and a theoretical basis for the definition and measurement of health and disability has been conceptualized. Cerebral palsy can be described across both concepts.

2.1.3 Measures of health and disability

The ability to evaluate individual care plans, service planning, interventions, and policies in children with cerebral palsy would inform on the quality of life of these children. Research on CP inAfrica has shown that a biomedical approach to disability is more common, and the International Classification and Functioning is not used as a guiding framework.(13) There is need to explore body function and structures which are descriptive in nature, whereas interventions can modify activities participation, environmental and personal factors in a child's life.(14) Therefore investigation into the measures of a child's health and disability most often are investigated based on these modifiable factors. The impacts of the illness or treatment on the child with CP, is hardly captured by the more traditional outcome measure which are directly related to the condition being treated, the symptoms and particularly survival. Inclusion of a more an all-inclusive outcome, such as health-related quality of life (HRQOL), is recommended.
For children with cerebral palsy (CP), quality of life (QOL) has been increasingly positioned as animportant outcome indicator.(15, 16)

There are no specific treatments for the resulting brain insult that lead to the motor dysfunction

ofCP. Instead, treatment is focused on a range of therapies with the aim of enhancing a patient's overall quality of life. In addition, interventions to reduce comorbidity usually does not eliminate

the intractable diagnosis, but could focus on reducing the impact of a childs impairment and potentially reduce the number of disabilities that accompany the musculoskeletal challenges of CP. Comorbidity however is strongly associated with psychosocial QOL.(17) Focusing on reducing CPcomorbidities could have a positive impact on psychosocial QOL. Children with CPs' psychosocialQOL has been shown to be reduced in spite of improvement in musculoskeletal and physical impairment.(17) Research indicates that paediatric and adolescent patients with CP have impaired functional and psychosocial QOL when compared with their normative peers,(18-20) This has led to a call for further examination of other patient level variables that could influence psychosocial QOL.(21)

QOL is generally conceptualized as a broad assessment of well-being across various domains and HRQOL and is considered to be a subdomain of the more global construct of QOL.(22) There are various generic measures for CP health related quality of life such as the KINDL, KIDSCREEN andChild Health Questionnaire. The choice of the PedsQL in this setting was due to its previous use in African settings and it consists of both the generic and the disease specific modules.

The paediatric Quality of Life (Peds QL) literature includes both generic and condition-specific instruments.(23, 24) The PedsQL 4.0 generic instrument is designed to be applicable to all population subgroups and is useful for comparing outcomes between populations. Condition specific instruments such as the PedsQL 3.0 CP module are designed to be applicable to one group and is useful in detecting outcomes arising from changes in this condition or factors associated withit. PedsQL assessments can be based on parent report, child reports or proxy

reports, or preferably acombination of all.(25)

2.1.4 Epidemiology of Cerebral palsy

Cerebral palsy is the most common neurologic and motor disability in children in the world including Africa.(26) Cerebral palsy (CP) describes a group of permanent disorders of the development of movement and posture causing activity limitation, which are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain.(27) It is an umbrella term which encompasses a range of different etiologies and phenotypes as well as being associated with a variety of comorbid conditions.(28) Cerebral palsy motor disorder, is often accompanied bydisturbances of sensation, perception, cognition, communication, and behaviour, epilepsy, secondary musculoskeletal problems and nutrition. There are insufficient community studies on cerebral palsy from the lower middle income region, however a recent population survey from Uganda describes the prevalence 1.8-2.3 /1000 children.(29) Bilateral spastic CP was commonest (45%); moderate impairment in gross motor function was present in 43%. Learning disability (75%) and epilepsy (45%) were the commonest co-morbidities. An earlier systematic review on CPin Africa reported the difficulty in obtaining accurate prevalence and clinical details on types and severity of impairment.(30) This is because the majority of articles on CP in Nigeria are from hospital based sources. (31) A recent Nigerian hospital based study report that two-thirds of the affected children have severe functional disability, and that there is a high default rate from rehabilitative care and only about one-third of affected children are given opportunities for any form of education at all with access to specialised education limited.(32)

Identification of children with CP within the community in low resource setting has been done using the key Informant Methodology, (33, 34) and more recently through a survey.(29) The Key Informant Methodology (KIM) has been shown to be effective for identifying children with

severevisual impairment and blindness and children with disabilities,(35, 36) including those with CP, inlower–middle income countries. The ten questions questionnaire has been validated in an African

community for use in the identification of children with neurodevelopment disorders including CP. Another tool for screening is the UN/Washington group questionnaire which is shorter and has a progression in the identification of functional limitation.(37) The KIM involves identifying recommended, respected community members who have had no previous medical training e.g., community leaders, teachers, local health or development workers. The key informants will undergo one day of training by the Principal Investigator using the ten question questionnaire as a screening tool and a picture booklet of children with CP.(37) After training, they will use a range ofapproaches to identify and list children who might have CP over a two-week period. Children they identify and their carers will be invited to a pre-agreed site, usually a primary health center, for comprehensive history and detailed examination, including neurological examination and confirmation of the diagnosis and classification of CP by a pediatric neurologist who would use specified diagnostic criteria. This involves a two-step screening exercise to identify children with CP.

2.1.5 Etiology and pathophysiology

Delayed diagnosis of CP in rural communities is common.(33) The most commonly reported causesof CP in developing countries include: birth asphyxia, kernicterus and neonatal infection (meningitis) with prematurity and low birth weight rarely.(31, 38) It is suspected, that the different causes of CP may affect the extent and type of associated co-morbidity including visual morbidity. For example, visuo-motor problems were seen to be greater in preterm children with CP than term children with CP.(39)

The prevalence of CP generally in high income countries is said be about 2.2/1000 children. A

recent study in Uganda which was a population based survey reported a higher prevalence of 2.8/1000 children. Most data on the frequency of CP is based on hospital data, as population surveys are expensive and rigorous to plan. Therefore, community studies using the key informant

methodology has been used in lower middle income countries and have yielded satisfactory results.However, estimates have been noted to be lower because of issues such as stigma in the case of CP.

Comorbid conditions are usually associated with severe presentations (quadriplegia and spasticity).This is more likely in Africa, where severe forms of the condition are relatively more prevalent.(40) Malnutrition specifically is a major problem in children with CP in lower middle income countries.(41) However, there are minimal data on the prevalence or management of these comorbidities in Africaand its impact on the education, daily living, social and emotional well-being of the child. A recenthospital based study showed epilepsy as the main comorbid condition that appears to encourage continuation with tertiary care.(32)

2.1.6 Classification, types of CP and severity measures

There are several classifications of CP. These are based on extensive research and their use helps to monitor trends in CP rate, to provide a basis for services planning and a framework for collaborative research. The Surveillance of Cerebral Palsy in Europe (SCPE), the most frequently used classification of CP, classifies CP as spastic, bilateral and unilateral, dystonia and choreoathetoid, ataxic CP and unclassified.(42) It facilitates comparison of results between differentstudies. In addition to this more medical classification, rehabilitation of children with CP has focused on the ability to function in their daily activities. Because of this, the examination of

children with CP is aimed at determining the functional level and abilities of these children as well as the type of CP.

Traditionally, the Gross Motor Function Classification System (GMFCS) is used to describe the severity of CP.(43) This is an evidence-based classification tool of five levels ranging from level I,which includes children with minimal or no disability with respect to community mobility, to level

V, which includes children who are totally dependent on external assistance for mobility. Restrictions in activity and participation has been described by the GMFCS which is used frequently in clinical practice and research.(44) The modification of the GMFSC-ER takes into recognition environmental modification and the use of assisted technology and environmental support.

A new framework for classifying manual function when children are handling objects in daily activities, is the Manual Ability Classification scale (MACS) of children and adolescents with CP.MACS demonstrates that it is a valid and reliable classification tool, although it is not as commonas the GMFCS to date.(45)

Another measure of function, is the Communication Function Classification Scale (CFCS). It is a standardized measure for assessing cognition. This involves an effective sender and receiver communication with both unfamiliar and familiar partners. Even though face to face communication, facial expression, gesture and body movements may be used in the test assessment,these signals may be difficult too for children with CP to understand. Parents most often shape theirconversations with children whose communication they find difficult to interpret, around signals that they believe the children can understand.

Children with motor disorders including CP, have been seen to take a respondent role in relationships and to use communication for a smaller range of functions than children without

motor disorders.(46, 47) The CFCS is a five-level classification system and the levels are determined based on the child's ability to communicate by using any method of communication usually used by the child and carer in a real life situation. It is a tool used for categorizing the communication ability of children with CP with familiar and unfamiliar partners.(48) The three functional classifications (GMFCS-E&R, CFCS and MACS) complement each other to provide abetter description of the functional profile of a child with CP.

2.1.7 Interventions for cerebral palsy

A multidisciplinary care approach is recommended for the medical management, development and to facilitate the educational potential of children with CP, if the child should achieve optimal results.(49) Even though guidelines already exist for the investigations and management of certain aspects of CP, little is known about the availability of guidelines or the recommendations for practice across resource-poor countries in Africa. Noted in Africa, is a lack of availability of adaptive equipment such as wheelchairs, wheelchair-accessible transport and other ambulation aids.These have been identified as barriers toward best care for children with CP in Africa.(50) In addition, limited access to health care facilities and specialists, inadequate knowledge about CP andassociated comorbidities in the environment has contributed to this obvious management gap.(40) High levels of social stigma towards children with neurologic disorders have been reported as reasons for families failing to seek treatment even when it was available.(51)

A systematic review on the impact of different interventions on quality of life (QoL) for children with cerebral palsy was conducted in 2010.(24, 52) This study investigated four key areas: (i) the clinical condition(ii) outcome measures: quality of life, well-being; (iii) study design: clinical trials;and (iv) target population. The results showed a positive effect from medicinal and motor control interventions on QoL. It was noticed however, that no single interventional approach could demonstrate a consistent positive impact on QoL across different studies or an overall impact.(24) Future studies investigating interventions are recommended.. This will help to (i) provide a clear definition of QoL, and investigate the relationship between symptoms' severity and QoL (ii) measure outcome at different time points to capture real effects of interventions and (iii) make moreuse of valid outcome instruments, either self-report or parent/caregiver proxy reports.

One of the 3 main strategies of the World Health Organization, Global Action Plan 2014-2019,

recommends that prevalence surveys of visual impairment and its causes should be conducted.(53)

Because CP is associated with a significant risk of visual impairment children with CP are a priorityfor targeted visual screening as well as the condition, cerebral visual impairment.(54)

2.1.8 Visual impairment in CP

Vision impairment is common in CP and, although it may be caused by problems with ocular structures, it is usually directly associated with the same brain injury which causes the motor problems, the 2 pathways being closely linked anatomically. This is called cerebral visual impairment (CVI) and can involve 'basic' vision such as visual acuity or visual field or 'higher' visual perception or cognitive vision such as the ability to see moving targets, to pick out a target ofinterest from a complex scene, visual control of body movement and object recognition. These cognitive visual problems may be termed perceptual visual dysfunction (PVD) and may be missed by conventional ophthalmological assessments.

Cerebral visual impairment is seen not only in children with CP but also with other neurodevelopmental problems such as epilepsy or hydrocephalus. Indeed, the declining prevalenceof treatable causes of childhood blindness such as corneal opacity, cataract and retinopathy of prematurity, and the success of paediatric intensive care survival rates, have led to CVI emerging as the commonest cause of impaired vision in children in developed countries. (55-57) In developed countries the commonest cause in children is from prematurity,(58, 59) in lower middle income settings, the commonest cause is from cerebral palsy.(60)

The clinical assessment of the visual system of a child with cerebral palsy is carried out for two main reasons: The first is to identify and manage treatable conditions such as refractive error, impaired accommodation and squint through an ophthalmological assessment which primarily focuses upon the structure of the eyes and the function of the eye including movements. The

second is to determine the functional vision, assessed with both eyes open, and behavior in order toidentify the visual limitations, and to plan how best to deal with them. Vision in a child is used for
access of information, visual guidance of limbs, social interaction and the overall development ofthe child, hence the importance of a functional vision assessment. The guiding definition of CVI used by most experts in the field of CVI includes: visual dysfunctions caused by damage to, or malfunctioning of the retrochiasmatic visual pathways in the absence of damage to the anterior visual pathways, or any major ocular disease, (61, 62) and more recently defined as a verifiable visual dysfunction which cannot be attributed to disorders of the anterior visual pathways or any potentially co-occurring ocular impairment.(63)

Assessment methods and treatments/interventions for CVI including the subgroup of PVD are lacking. CVI, particularly the subtype of PVD may be asymptomatic or unrecognized as many brain functions (such as visual guidance of movement and visual search) are subconscious, so that ifpart of the brain responsible for a specific function is damaged (especially from birth/infancy), theremay be lack of insight for the loss called agnosia.(64) Because children do not know and are unaware of the nature and degree of their visual problem they face, the condition is anosagnosia.(65) Therefore, they are not by themselves in a position to compensate for their visual difficulties without appropriate assistance. (47)

2.1.9 Terminologies in CVI

The terminology within the field of CVI is not uniform. It is an umbrella term analogous to CP. There is a spectrum of disorders under the term CVI. The term cortical visual impairment which is commonly used, describes the profound visual loss which primarily involves the primary visual cortex, (66) but has largely been replaced by the term cerebral visual impairment because the whitematter is often involved.

The term perceptual visual disorder relates to pathology higher up the visual pathways in the brain (visual association areas) where vision is ‘used’ to navigate through a complex visual seen or tosubconsciously control hand or foot movement, or simply to recognize something or someone.
Problems in these areas manifest in visual behavioral difficulties,(67-69). The terminology PVD isrelatively new and has yet to be fully defined and included in the ICD classification. It is however implicitly included in the International Classification of Functioning (ICF) aimed at according optimal service provision.

2.1.10 The perceptual System

We engage and access our physical and social environment learn through perception.(70) Perception is essential for knowledge acquisition, in both short and long term experiences and for the guidance and control of our behaviour and movements. There are different perceptual systems, they include: vision, auditory, smell, somatosensory and body perceptions. Perception involves allactivities in daily living that pertains to the recording, processing and coding of information, including cognitive, motor (eye, hand, limb and body movements) and emotion. Visual perceptual ability is classified as lower or higher visual capacities.(71) Lower or elementary visual capacities are described by: visual acuity and spatial contract sensitivity, the visual field, visual light and darkadaptation, colour vision, and spatial vision (localization, distance and direction perception, monocular and binocular depth perception, including stereopsis). Higher or more complex visual capacities include visual identification and recognition, topological and geographical orientation, text and number processing. In addition, moment to moment visual mapping of the surrounding environment which provides both the template for visual search and the fundamental means of bringing about guidance of movement through our surrounding.

2.1.11 Development of the perceptual visual system

The visual system develop has two peaks of development, mainly in the first 2 years and between 8-9 years, while normal development of basic social function develops during the first 3 years.(72,

73) The developing brain, possess its maximum plasticity in early childhood, therefore it is suspected that there may be a time frame within which interventions would yield bests results. PVDtends to become progressively apparent with age, the more severe features being possible to assess by 4-5 years.(74, 75) Therefore, visual follow-up to seek such dysfunction is required into the earlyschool years, especially when first attending school in order to identify features such as being unable to distinguish a friend or relative from a group. Visual acuities may be normal and associated visual field affectation may go unnoticed.(76)

2.1.12 Perceptual disorders

For normal development of perception to occur, cognitive, motor, motivational and emotional development all also need to occur and feed in. (77) Perception does not operate in isolation, but is entrenched in a network of mental functions, which include: attention, memory, executive function,action (eye movements, hand movements- drawing, writing, walking), curiosity (motivation) and mood.(66) These have an impact on perception but are also reciprocally influenced by perception. These reciprocal interactions mean that disorders of perceptions are not only caused by direct disturbance of a specific perceptual system but can also result as a consequence of dysfunction of the other functional systems (mental functions). Behavior is a final common pathway product whichis the result of a combination of cognitive, motivational and

social functions and represents the interplay of different functional systems. The diagnostic classification of such typical visual behavioral abnormalities sometimes face a major challenge, especially with similar conditions such as autism disorders and delayed developmental milestones. This is particularly because of the complex interaction of mental functions. The resulting impairments seen in children with CVI are therefore to be expected in more than one domain.

2.1.13 The dorsal stream

Anatomically, the dorsal stream connects the occipital lobes to three brain areas: the posterior parietal lobes (which process the visual picture and attention to specific aspects of the picture), the motor cortex (which allows movement through visual space) and indirectly to the frontal cortex including the frontal eye fields (which allows attention to be paid to specific aspects of the scene, by generating rapid head and eye movements to specific aspects of the scene). Investigation by Dutton et al, shows that bilateral focal damage affecting the dorsal stream may give rise to a varietyof presentations, commonly: Impaired visual guidance of movement (known as optic ataxia), diminished capacity to both process as many items within the visual scene as normal, and to chooseto give attention to them (Attention cannot be given to items that are ‘not there’ owing to a diminished attentional substrate capacity of damaged posterior parietal lobes leading to saturation of the system), bumping into items that have not been processed, diminished substrate for visual search, lessened capacity to give simultaneous attention to data coming in through the different senses, leading to one sense appearing to compete for another and diminished capacity to identify the location of sound.(78)

2.1.14 The ventral stream

The ventral pathway runs from the occipital lobe to the occipitotemporal and temporal lobes on

each side of the brain.(68, 79) The temporal lobes sub serves, object recognition and visual memoryas well as being responsible for providing a rich and detailed representation of the world. They enable recognition of objects and faces, accurate orientation and navigation by means of recognition, and a sense of direction.(80-82) It has been noticed that dorsal stream dysfunction can occur in isolation or with additional ventral stream dysfunction, but (64, 83) Isolated ventral streamdysfunction is rare.

2.1.15 Classification and tools used to identify CVI

CVI has been classified broadly in 3 levels of severity. ((64, 84). The first group are those with profound visual impairment typically referred to as cortical blindness affecting the primary visualprocessing areas. The second group, have significant visual impairment which may be associatedwith cerebral palsy and intellectual and attentional disorders affecting both the motor processing areas and secondary associated visual processing areas. The third and most mildly affected grouphave normal visual acuity but perceptual visual disorders, where only the secondary associated visual processing areas of both ventral and dorsal streams or dorsal stream alone are involved (PVD)(74). Thus the term CVI includes a spectrum of deficits which may range in severity from mild to profound and may occur in combination.(66)

Recognizing PVD is an important step toward habilitation, even though difficult in some circumstances. Conventional acuity measurements do not give the complete picture(85) Perceptual visual impairment or dysfunction can be suspected from observed visual behaviors of a child by thecarers, reported in careful structured history taking.(86)

Structured history taking potentially formed the basis for the development of this standardized approach for eliciting typical behaviors of children with PVD. This was started by the sets of clinical history questions devised by Ferziger et al. and by McCulloch et al. to elicit information about the visual skills of children with severe cerebral visual impairment (110). This structured history from parents / carers provides the most efficient and comprehensive means for identifying,screening and characterizing many aspects of the behavioral characteristics and adaptations to PVD.(87-89) It is suggested as the most efficient means of screening for and making a preliminaryindication of CVI/PVD and has been validated in the UK.(90)Compared to formal psychological testing which may be hindered by visual disorders and other disabilities.

This identifies specific behavior differences from that of a normal child who does not have visual dysfunction in children. Most importantly the inventory can be used to assess what kindof services the child requires and specific tailored recommendation for environmental and behavioural modifications can be made.(91) Furthermore, the questions are also useful to guide parents in their assessment of the child's behaviour and for education purposes. A team of researchers from the UK have worked on the 'Insight' structured clinical question inventories to elicit symptoms of perceptual visual dysfunction in brain injured children.(92) They utilized a structured (closed set of questions) history taking, involving the development of clinical history inventories called the Insight history questions inventory (Appendix 1).

World Health Organization, through the International Classification of Functioning, Disability and Health (ICF), clarified the understanding of CP in relation to intervention options and differentiated functioning problems, participation problems and disability. It is suggested that interventions shouldaim at maximizing a child's independence in daily activities and community participation, while also focusing on optimizing children's environment. In addition, a goal-based approach and (66), patient centeredness based on choice of interventions guided by what would best help the family achieve their goals, is recommended.(93)

Visual impairment in children with CP varies in nature and severity and its prevalence ranges from40-50% of children in different studies (60). Visual impairment in CP is often directly associated with the same brain injury which causes the motor problems. This is generally termed cerebral visual impairment (CVI) (84), commonly affecting children with CP but also children with other neurodevelopmental diagnoses such as epilepsy and hydrocephalus. CVI can cause problems with 'basic' vision such as visual acuity or visual field or affect 'higher' visual perception or cognitive vision such as ability to see moving targets, to pick out a target of interest from a complex scene, visual control of body movement or object recognition (90). This

latter group of behavioural
symptoms of abnormal higher-order visual processing can be termed perceptual visual dysfunction(PVD), part of the CVI spectrum.

It was previously shown in India, Bangladesh and more recently in the United Kingdom,(91, 92, 94), that this latter group of symptoms of higher visual processing problems or PVD are commonlypresent in children with CP; we have previously published a detailed visual assessment of the children recruited for this trial showing high rates of visual pathology (including 46% PVD measured objectively and 49% CVI (dropping to 16% if optic atrophy excluded) (95).

Previous work has suggested that PVD can be effectively assessed by a structured clinical history question inventories, including the insight question inventory (IQI) (91, 92, 96, 97). IQI scores havebeen shown to have internal reliability, and to discriminate between children diagnosed with CVI and healthy aged matched volunteers (68, 96); they have also been shown to correlate with neuropsychological tests of visual perception (92), and predict quality of life in children with CP independent of other predictors such as visual acuity and degree of motor impairment (91).

The IQI provides in-depth information about the aspects of daily living activities that children struggle with. The current 52 item inventory, Insight Questions Inventory tests 6 domains of vision namely, visual field, perception of movement, visual guidance of movement, visual search, visual attention, recognition and navigation, which, in addition to visual field, test visual perception, eitherdorsal (occipito-parietal) stream processing (visuo-motor control, processing moving targets and processing large amounts of visual information at once) or ventral (occipito-temporal) stream processing (person and object recognition) (3).

Previous work indicated that considering dorsal and ventral stream as 2 factors explained 63% ofvariance of IQI scores between patients.(94) In addition to diagnostic information, a simple

software program links each question inventory response to a specific group of tailored visual support strategies (VSS) appropriate to that question, so that after completing the inventory each

child/family has a set of tailored visual support strategies (IQI VSS) for that particular child. An example of the IQI and Visual support strategy can be seen in question 11, which asks “Does yourchild bump into door frames or partly open doors (left/right/both)”? With corresponding tailored visual support strategy response from caregivers who would give extra hints. For example, "Thereis a door coming up in a few steps." Another recommendation would be to replace doors with a beaded curtain.

A recent hospital based longitudinal study investigated the impact of the IQI VSS linked to the inventory responses on functional vision and quality of life. (92) Children were followed up 6 months after receiving the IQI VSS and improvements were seen in both qualities of life and functional vision compared with baseline pre-intervention assessments but there was no control group. Our previous work has suggested that PVD (measured by IQI) adversely affects quality of life,(91) and that IQI VSS improve quality of life (92), using the PedsQL 4.0 Generic module which assesses domains of physical activities, social, emotional and school functions. The PedsQL

3.0 CP module, is designed to be used by children with CP to detect changes arising from this condition or factors associated with it, with subdomains which include daily activities, movement and balance, fatigue, pain, school functions, eating and speech (23, 98, 99). Peds QL 4.0 Generic and PedsQL 3.0 CP assessments can be based on parent report, child reports or proxy reports (25, 100). SincePedsQL3.0 CP is specifically designed for children with CP but has not been previouslytested in relation to CVI or PVD we decided to use both PedsQL 4.0 Generic and PedsQL 3.0 CP modules as primary outcomes for this trial.

In the development, the Insight questions inventory was approached initially as a questionnaire which went through a series of validation process. The current 50 question item inventory, tests 7domains of vision namely, visual field, perception of movement, visual guidance of movement, visual search, visual attention, recognition and navigation, and abnormal behaviours in crowded
places. This further describes mainly dorsal stream processing (visuo-motor control, processing moving targets and processing large amounts of visual information at once) or mainly ventral stream processing (person and object recognition) or both. This describes and estimates the locationand extent of the brain damage clinically.

The utility of the IQI has been investigated in a number of different settings.

Specifically, it has been shown to be internally reliable when tested on a group of children with CVIdiagnosed by an expert and compared to age matched typically developing children from the general public (96). Potential utility as a screening/diagnostic tool in a vulnerable population of children was suggested in a study of ex-premature children who had all passed neurodevelopmental screening. Around a third gave high rates of positive answers i.e. they reported significant problems with visual perception in everyday life. When given objective tests of visual perception and compared to a control group, the premature children scored significantly lower and this difference was shown to be accounted for by the children who had responded positively to the IQI.(96, 101) Further evidence of correlation between IQI response scores and formal neuropsychological tests ofvisual perception was demonstrated in another UK hospital based study of children with all cause CVI. (92) So there is some evidence linking subjective PVD as reported by IQI and objective PVD as measured by a range of psychophysical tests. there is also evidence of correlation between IQI scores and quality of life scores. In a group of children with CP in rural Bangladesh, IQI scores predicted poorer quality of life independent of

visual acuity impairment and motor impairment. (91)

Another way of assessing / classifying visual perceptual deficits involves using formal neuropsychological tests of visual perception.(84) The need to be administered by a qualified clinical psychologist which limits accessibility even in high income countries and may not be practical in LMICs. Therefore, current research suggests that the structured inventory approach maybe worthy of further testing in LMIC settings.

Another potential advantage of the structured inventory approach is that, on the basis of behaviouralpatterns elicited, appropriate specific adaptive habilitational strategies (or visual support strategies, VSS) can be applied and delivered (Appendix 2). The purpose of the VSS is to adapt behaviour or modify the environment to allow for the specific visual perceptual problem reported and minimize its impact on mobility, communication, development and education in the life of the child. The ultimate intention is to accomplish maximal independence into a productive and meaning full adult life. A software program links question inventory response to the appropriate visual support strategies so that after completing the inventory each child/family has a tailored set of strategies for that particular child (a sample report is seen in Appendix 3. A small uncontrolled pilot study showedthis approach to be effective in improving quality of life in a hospital based series of all cause CVI in London.(92)

It is important to mention that the IQI is not a screening tool but part of a detailed clinical assessment that leads to a clinical impression of which streams of visual processing are most affected and to a set of tailored adaptive visual support strategies linked to the specific responsesgiven by the carer. A 5 question version of IQI has been piloted as a screening tool.(54)

In order for the Inventory to be used in international clinics, as well as, a valid research instrumentin international clinical trials, it must be culturally adapted and the translations need to be linguistically validated for the targeted population.

2.1.16 World Health Organization (WHO) definition of visual impairment

The WHO definition for visual impairment refers to impairment of the basic vision with reference to visual acuity (It is the spatial resolving capacity of the visual system and measures the sharpnessof vision. The International Classification of Diseases 11 (2018) classifies vision impairment into two groups, distance and near presenting vision impairment.

Distance vision impairment: Mild –visual acuity worse than 6/12 to 6/18, Moderate –visual acuity worse than 6/18 to 6/60, Severe –visual acuity worse than 6/60 to 3/60 and Blindness – visual acuityworse than 3/60. While for near vision impairment, near visual acuity worse than N6 or M.08 at 40cm is defined as near vision impairment.

2.1.17 Clinical visual assessment

The performance of components of the visual system are assessed by vision related abilities ad arereferred to as visual function while functional vision are assessed by visual task-related ability.(102) behavioral methods and test designs that have been developed to measure what we consider pillars of visual function. These visual function outcomes include: visual acuity, contrast sensitivity, color, depth, and motion and lastly, visual fields.

How an individual uses their vision in everyday life is not only known through assessment of visualfunction. Certain tests may be useful for assessing and characterizing vision in individuals with ocular based conditions, however, they may be less informative in children with brain based visual impairment such as in CVI. This may be seen when considering individuals with CVI who may be normal or near-normal in performance on assessments of acuity, color, and contrast, yet these sameindividuals will often describe a variety of visual perceptual deficits such as difficulties with visuo- spatial/motion processing, environmental complexity/crowding, and sustaining attention during tasks with high visual demands. (62)

2.1.18 Patient centered care

The use of targeted, tailored strategies in the management of CVI is in line with the concept of patient centered care. Ensuring that families and care givers are involved in, and at the center to their care and choices, is now recognized as a key component of developing high quality healthcareand obtaining better outcomes.(103) The Insight questions inventory and tailored VSS provides the

use of a checklist and offers interventions which are customized to each child and reflects patient needs, values and choices. The Insight history questions inventory and visual support strategies,

models a pathway through which a patient-centered intervention can contribute to better outcomes. The VSS are designed to be practiced by the caregivers at home and school. The IQI tool can be administered by any health professionals trained to do it but this should be done in conjunction witha comprehensive baseline visual/ophthalmological assessment.

2.1.19 The impact of failing to make a diagnosis

The impact of PVD on the life of a child may be far reaching. Failure to or an incorrect diagnosis can lead to a wrong diagnosis such as delayed visual maturation, intellectual impairment, autism, ordescribed as a child with bad behavior, aside from having a poor performance in school.(83, 101) These negative impact of PVD can lead to lack of confidence, self-worth and anger, while the failure to implement adequate visual support strategies may impairs learning, with a potentially greater measure of poor social interaction, communication and dependency on careers and family.

2.1.20 Quality of life in children with cerebral palsy

Quality of life (QoL) is related to disability.(104) QoLis a complex multidimensional concept including physical health, psychological and social well-being and beliefs. QoL and health status measures are largely subjective and can be determined through the judgment of individual and otherpeople about their own health and life status. Studies have been conducted to evaluate the factors that influence the quality of life in children with disability and disease conditions.

Childhood disabilities are conditions that do, or are highly likely to, affect the trajectories of children's development into adulthood. Many of these have a neurological basis. Often, additional

impairments are seen to be part of the neurologic condition. Examples of complex conditions withcorrespondingly complex disability include, musculoskeletal conditions or genetic syndromes, cognitive, behavioural and communication disorders. (13)

The World Health Organization (WHO) proposed a new, more dynamic and empowering definitionof health: ‘health is the ability to adapt and to self-manage’(105) The International Classification ofFunctioning, Health and Disabilities framework has been influenced by this new thinking is and involves 6 main areas. Functioning, family, fitness, fun, friends and future. The idea is that thinkingabout these F words at clinical level would promote personalized interventions.(13)

Measures of outcome of intervention involving children with CP, are expected beyond medical outcomes and include the child’s ability to communication, coping, and problem-solving skills. Interventions are expected to impact on the educational opportunities, family adaptation and the useof services. Quality of life (QoL) is therefore an important addition to standard biomedical reports and should be encouraged as a measure of clinical progress and research outcome.(23, 106, 107)

Peds QL is a modular instrument designed to measure HRQoL in children and adolescents 2 to 18 years old. It is has a generic core integrated with a disease-specific core.(98) The PedsQL assessment tool, is used in African countries including Nigeria in the study of children with CP.(108) The instruments are contained as a child self-report and parent proxy-report formats. The Parent proxy-report includes ages 2–4 (toddler), 5–7 (young child), 8–12 (child), and 13–18 (adolescent), and assesses parent's perceptions of their child's HRQOL. The face content for each ofthe forms are essentially identical, the difference between the instruments based on age is in the useof age appropriate language, and first or third person tense. The problems in the last one month are focused on in the questions of the item to be answered. A 5-point response scale is utilized across child self-report for ages 8–18 and parent proxy-report (0 = never a problem; 1 =

almost never a problem; 2 = sometimes a problem; 3 = often a problem; 4 = almost always a problem). Items are

reverse-scored and linearly transformed to a 0–100 scale (0 = 100, 1 = 75, 2 = 50, 3 = 25, 4 = 0), so

that higher scores indicate better HRQOL. Scale Scores are computed as the sum of the items divided by the number of items answered (this accounts for missing data). The Scale Score is notcomputed if more than 50% of the items in the scale are missing.

The two modules proposed for our study are the PedsQL 4.0 generic and the PedsQL 3.0 cerebralpalsy modules. The PedQL Generic module has 4 subscales which identifies problems with: 1) Physical functioning, 2) Emotional Functioning, 3) Social functioning and 4) School functioning.The PedQL-CP module has 7 subscales which identifies problems in activities in everyday living,they are:1) daily activities 2) School activities, 3) Movement and Balance, 4) Pain and Hurt, 5) Fatigue, 6) Eating activity, 7) Speech and communication.

2.1.21 Current research gaps and CVI research focus

After finding that the IQI/VSS approach may contribute to assessment and management of childrenwith CP and or CVI in the UK, Bangladesh and India it is desirable to test and adapt its use in an African population.

2.2 Hypotheses

The research is based on the following underlying hypotheses:

1. Structured history taking from parents and careers (in addition to standard ophthalmological assessment) using standardized clinical question inventories is a useful way of determining achild's perceptual visual dysfunction (PVD) in the presence of brain injury.
2. Practical visual support strategies aimed at adapting specific aspects of the child's environmentor behaviour in the light of the responses to the question inventory may improve children's quality of life

3 Study design

3.1 Aims

The overall aim of this study is to improve management and quality of life of “school-aged childrenwith cerebral palsy through assessing and adapting to their visual co-morbidity

Objectives

Phase 1

1. To determine the effectiveness on the QoL and visual function of children with perceptual visualdysfunction in children with cerebral palsy following the application of specifically targeted Insight visual support strategies (IQI VSS), compared to general supportive management: Protocol for a randomized control trial (Chapter 4)
2. To investigate if the current Insight question inventory and associated recommended visual support strategies are culturally relevant and acceptable, for use in children with cerebral palsy inCross River State, Nigeria and whether it can be suitably culturally modified. (Chapter 5)

Phase 2

1. To determine the etiology, prevalence, pattern and characteristics (including co-morbidities) ofchildren with Cerebral Palsy in Cross River State Nigeria. (chapter 6 and 7)
2. To determine the prevalence, pattern, and characteristics of visual impairment including perceptual visual dysfunction in children with Cerebral Palsy in Cross River State Nigeria.(chapter 8)
3. To determine the effectiveness of the IQI VSS (compared to general supportive management) onthe QoL and visual function of children with perceptual visual dysfunction in children with cerebral palsy following the application of specifically targeted Insight visual support strategies, (chapter 9)

Study Site

This study was carried out in the 3 senatorial districts consisting of 18 local government areas of Cross River State Nigeria (Figures 1 and 2) which has a population of 3.5 million. There is a Federal University Teaching Hospital which has a center that offers tertiary care eye services for children. The University Teaching Hospital has 15 medical subspecialties including a Pediatric Neurology unit in the Department of Pediatrics, and a Physiotherapy Department.

Cross River state is located in the South region of Nigeria. It has a population of 3.738 million (2016 population projection) with an areaof 21,019Km^2. There are 50.9% males and 49.1% females. Children 0-14 years make up 38.2% of the population. The state is mainly a civil service state with indigenes fishing and farming. Compared with other states in Nigeria, it is one of the poorest states in the country, however itposes one of the best primary health development agencies. Compared to other states in the country, the state has poor Health indicators. Maternal mortality rate of 2000 deaths per 100,000 live births, under five mortality rate of 176 deaths per 1,000 live births and an infant mortality rate of 120 deaths per 1,000 live births (109) Compared to other African countries, Nigeria has one of the worst health indices especially for under five mortality and infant mortality rates by the world bank report of 2019.

Currently Nigeria has 600 Paediatrics for a population of 40 million children, 40 Paediatric neurologists, and 18 Paediatric Ophthalmologists in the country. The management of children with cerebral palsy is poor as there is a dearth of human resources to diagnosis CP early and easily andoffer the much needed habilitation services.

In Cross River State, there are four major languages: French, Efik, Bekwarra, and Ejagham, however, the common language used forcommunication between communities across the state is a variant of English language.

Figure 3.1. Map of Nigeria showing the location of Cross River State

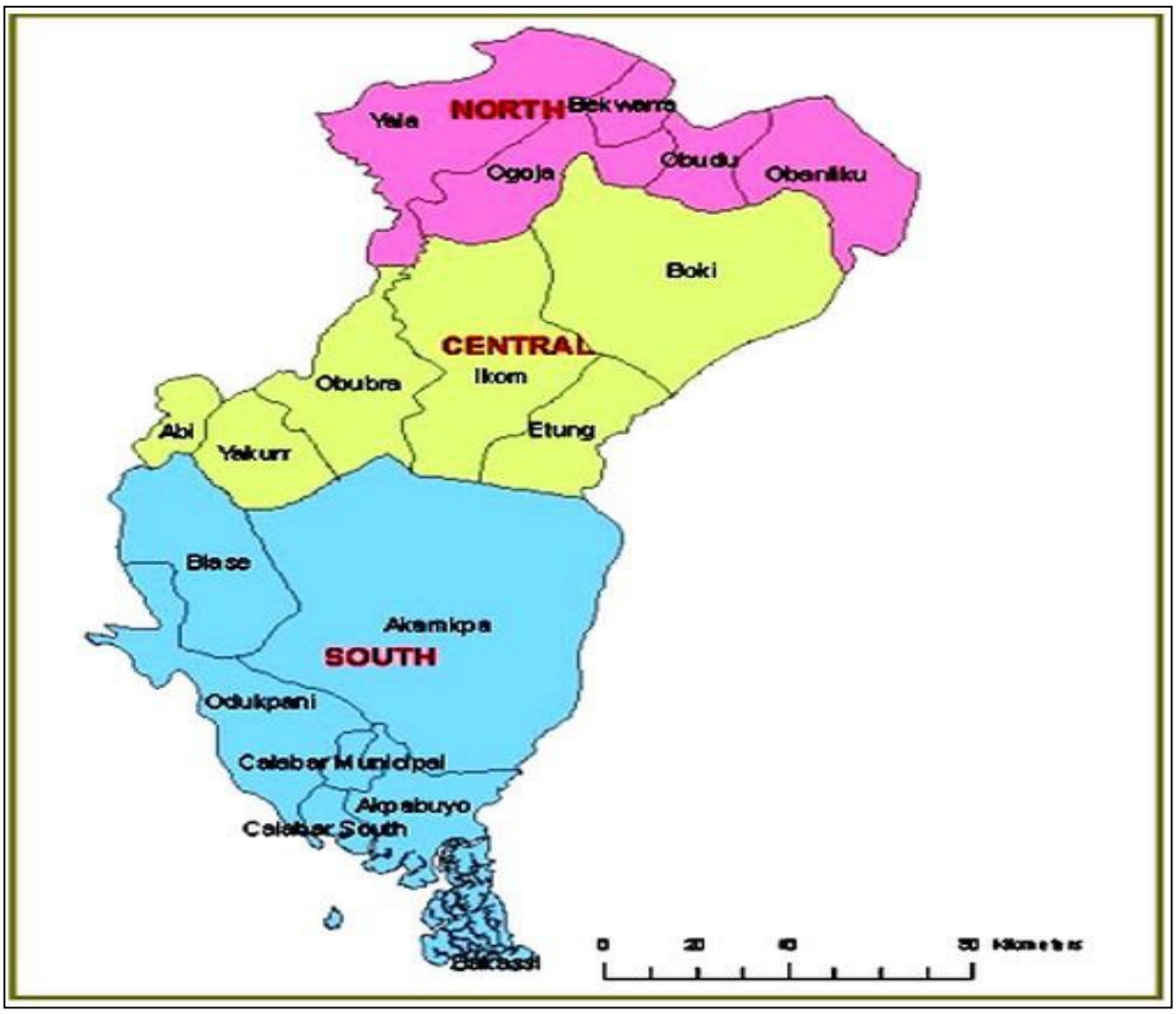


Figure 3.2. Map of Cross River State, the senatorial districts and local government areas.

1.1 Study methods

3.2.1 Phase 1

1. Translation and cultural adaptation

Piloting/adapting of the Insight question inventory and strategy for use in Cross River State population.

The current British version of the IQI was created for an English community with references to terms and expression and content. The IQI therefore needed to have to local reflections and expression to the IQI tool. In order to obtain a tool that was going to be understood by the Cross River community it was necessary for the British IQI to undergo\ translation and cultural adaption. Preparation for the translation into an acceptable and understood English version was work carried out before the translation work began by the principal researcher, by reviewing the English version of the IQI. A forward translation (cultural and linguistic adaptation) of the original IQI document (source), versionof the instrument into the Nigeria version, was performed by 2 independent translators. Reconciliation—comparing and merging of the versions produced by the 2 interdependent scientists (sociologist and linguistic specialist) into a single forward translation gave rise to the Back translation which was performed by a language specialist who compared the new version with the original British version, to highlight and investigate discrepancies between the original and the reconciled translation. Cognitive debriefing was performed first in a Pediatric neurology, clinic testing the instrument on a small group of relevant patients or lay people in order to test alternative wording and to check understand ability, interpretation, and cultural relevance of the translation; The review of the cognitive debriefing results and finalization—comparison of the patients' or lay persons' interpretation of the translation with the original version to highlight and amend discrepancies was performed, followed by a community field study using parents in the community with children who have CP to assess the translation and cultural adaptation.The final report produced the Nigerian version of

IQI which was used in a larger clinical trial.

Adaptation of the IQI to local setting – 2 steps (International Test Commission Guidelines on Translating and Adapting Tests for translation/adaptation of existing tests and instruments).(110) The purpose of the test adaptation of the IQI included the following: To check whether or not the test in a second Translating and Adapting Tests including the language and culture could measure the same construct in the original version of the IQI. Translators were selected; choosing a design for evaluating the work of test translators (e.g., forward and backward translations); choosing any necessary accommodations; modifying the test format; conducting the translation; checking the equivalence of the test in the second language and culture and conducting other necessary validity studies. Cognitive debriefing was used to actively test representatives of the target population and target language group to determine if the respondents understand the questionnaire the same as the original would be understood. It was also used to test the level of comprehension or understanding of a translation by the target audience, and/orto test alternative translations and was used to determines if translations would be deemed inappropriate or confusing by the target population.(111)

Therefore, the following steps were taken:

a. Forward translation (independently by social scientist and linguist) – v1

b. Face validation and comparison of version 1 by reconciliation committee – v2

c. back translation by second language specialist – v 3

2. Adaptation of questions in response to feedback from parent/carers

a. review of v3, clinic testing and cognitive debriefing (n=8 of parents recruited from the neurology clinic, who would not use theIQI in future; by the research assistant through purposive sampling – v4

b. IQI administered (n=24); Understanding of items in the IQI checked in community recruited sample of parents n=24 with cognitive debriefing using categorical scale of understanding

:(P1); no difficulty understanding or remembering the question; (P2)a difficulty remembering the question; (P3) a different understanding of what the question referred to; (P4) the respondent had difficulty recalling, formulating or reporting answer (P1-P4) assessed for each of the 52 questions in the IQI – v4

c. exploration of parent/carers understanding of the IQI (using categorical measure of understanding) – v5 (used in the RandomizedControlled Trial)

3.2.2 Phase 2

1. Cross sectional study

   A cross sectional study was conducted using the key informant methodology to identify children with CP in the communities.

2. Randomized control trial

   A randomised control trial of the Insight Questions Inventory and visual support strategies against standard care for the management of CP wasconducted to investigate impact on quality of life of children with CP.

Effect of visual support strategies on the quality of life of children with cerebral palsy and cerebral visual impairment / perceptual visual dysfunction in Nigeria: study protocol for a randomized controlled trial

Authors

* Roseline Duke 1,2, Komomo Eyong3, Kathryn Burton4, David MacLeod2, Gordon N Dutton5, Clare Gilbert1, Richard Bowman1

1. London School of Hygiene and Tropical Medicine, International Centre for Eye Health, London WC1E 7HT, United Kindgom.

2. Calabar Childrens' Eye Center, University of Calabar Teaching Hospital, Department of Ophthalmology, Calabar, Cross River State, Nigeria dr.roselineduke@gmail.com

3. Paediatric Neurology Unit, University of Calabar Teaching Hospital, Department of Paediatrics, Calabar, Cross River State, Nigeria. komomoeyong@yahoo.com

4. Cambridge Community Services NHS Trust, United Kingdom kjburton@doctors.org.uk

5. 45 Station Road, Milngavie, Glasgow G62 8BS dutton@duttonhome.com

6. Clare.Gilbert@lshtm.ac.uk

7. Richard.Bowman@lshtm.ac.uk

Corresponding Author: *Dr Roseline Duke

London School of Hygiene and Tropical Medicine, London WC1E 7HT, United Kingdom

Calabar Childrens' Eye Center, University of Calabar Teaching Hospital,

Department of Ophthalmology, Calabar, Cross River StateEmail:

roselineduke@unical.edu.ng

4.1 Abstract

Background

Cerebral visual impairment, including perceptual visual dysfunction (CVI/PVD), is common in children with cerebral palsy (CP). Inventories of questions relating to practical aspects of visual perception in everyday life, in particular the closed ended Insight Questions Inventory (IQI) can be used to assess CVI/PVD. Linking responses to the inventory with specific visual support strategies, aimed at modifying the child's environment and or behaviour to minimize the impact of the CVI/PVD has been piloted. The IQI and tailored strategies have not been used in an African population, nor has it been tested in a controlled trial. This trial will compare the effectiveness of the IQI and linked visual support strategies versus general supportive treatments on the quality of life of children with CVI/PVD in CP through a randomized controlled trial (RCT).

Methods/Design

This is a prospective, double blind, parallel-arm, randomized controlled trial. The primary outcome is change in quality of life scores between the two arms of the trial at 6 weeks, assessed using the Paediatric Quality of Life Inventory (PedsQL) CP 3.0 module. All children will undergobaseline assessment including: Open questions inventory, IQI, PedsQL 3.0,

PedsQL 4.0 generic, and Strength and Difficulties Questionnaire.

Eligible children with CP aged 4-<16 years will be stratified and blocked by age group 4-9 and 10-<16 years and by Gross Motor Function Classification System (GMFCS) Levels 1-3 and 4-5). Families in the intervention arm will receive tailored Insight visual support strategies and phone calls during the six-week trial period. The control arm will not receive the intervention until after the six week trial period. Follow up interviews will be performed in both arms at six weeks with a repeat administration of the PedsQL CP 4.0 and PedsQL generic 3.0, the IQI and the strength and difficulties questionnaire. Secondary outcomes include a change in functional vision, measured by the insight questions inventory, between baseline and six weeks.

Conclusion

This will be the first randomized controlled trial for any visual intervention for children with CP despite the fact that it is known that there is a high risk of visual impairment in such children. This study compares the use of tailored (Insight) visual support strategies with current treatment for the management of children with CP who have PVD in a resource poor setting, and will provide evidence of the effectiveness of this intervention.

Trial Registration: Pan African Clinical Trials Registration Number: PACTR201612001886396. Registered on the 3rd December 2016. URL oftrial registry record: Registry URL: http://www.pactr.org/ PACTR201612001886396

Keywords: cerebral palsy, cerebral visual impairment, perceptual visual dysfunction, Insight Questions Inventory, visual support strategies,paediatric quality of life

4.2 Introduction

Cerebral palsy (CP) is the most common cause of motor disability in children in low income countries, affecting the wellbeing of children and carers.(40, 112) It has a worldwide incidence of about 2-2.5 per 1000 live births .(31, 40, 113) Cerebral palsy describes a group of permanent developmental disorders of movement and posture causing activity limitation, which are attributed to non-progressive disturbances in the developing foetal or infant brain. In most individuals with CP the motor impairment is accompanied by secondary musculoskeletal problems, epilepsy, disturbances of sensation, perception, cognition, communication, and behaviour.(28, 114, 115) The definition of CP highlights that themotor and visual impairments in CP are integral components of the same disorder.(28)

The diagnosis of CP is based on clinical assessment from history and examination.(116) For clinical and research purposes CP is classified by the nature of the movement disorder (spasticity, ataxia, dystonia, and athetosis), and the anatomic distribution of the motor abnormalities.(42) Current standardized classifications of CP include the Gross Motor Function Classification System (GMFCS), the GMFCS expanded and revised(E&R), the Manual Ability Classification System (MACS), and the Communication Function Classification Scales (CFCS). There is currently no scale assessing visual or sensory function.

Facility based studies of children with CP in Africa have shown high rates of the more severe forms (using GMFCS) of CP and of associatedconditions,(34, 117) such as epilepsy, learning disability, deafness, speech disorders, visual impairment and malnutrition.(118)

The development of normal vision takes place over the first few years of life as anatomical structures and physiological processes mature.(119)This occurs at the same time as the development of language, emotion, motivation, mental functions which includes cognition, social functionsand the motor systems, which means that poor visual function can affect these

functions and vice versa. Behaviour mirrors the cooperation andclose integration of these functional systems.(28, 120) Mental and behavioural difficulties can be assessed using the Strengths and Difficulties Questionnaire (SDQ), a screening tool for 3-16 year olds (not specific to CP). It has 25 items on psychological attributes across 5 domains: emotional symptoms, conduct problems, hyperactivity/inattention, peer relationship and pro-social behaviour. (121)

Cerebral visual impairment (CVI), the commonest form of VI in CP. (60, 122) refers to loss of visual function from lesions in pathways from thelateral geniculate body and/or from the cortex to the higher centres. The latter, known as the perceptual visual system of the brain, includes the dorsal (occipito-parietal) and the ventral stream functions (occipito-temporal). (78, 123) Lesions between the chiasm and cortex can lead to loss of visual acuity and/or defects in the visual fields. Lesions in the perceptual visual systems give rise to perceptual visual disorders (PVD) such as problems with face recognition, finding objects in a complex visual environment, or orientation in space. These have received little attention compared to loss of acuity or visual field but studies have shown that PVD can impact on a child's ability to function using vision and adversely affects quality of life.(82, 91, 124) The deficits in CVI/PVD may be mild or severe, ranging from mild PVD with normal visual acuity and visualfields, through to complete loss of all visual function, depending on the site(s) and severity of the lesion(s). The term CVI/PVD is used to reflect the spectrum of disorders. Cerebral visual impairment/PVD affected children may also have intellectual and attentional disorders. As many brain

functions are subconscious, such as visual guidance of movement and visual search, CVI/PVD may be asymptomatic or go unrecognised. Inaddition, in children with CP motor deficits can mask disordered visual guidance of movement.(82)

Cerebral visual impairment/PVD can be suspected from observed visual behaviours, and becomes more apparent with age. The more severe features can be assessed by 4-5 years.(75)

Children who do not have sufficient language or who are unaware of their condition, cannot report or comment on what they can see or why vision is problematic, and therefore, behaviour based methods are used for assessment.(62, 86, 125) Although formal psychological testing may be helpful in identifying children with CVI/PVD,(87) appropriateness of psychological tests in a clinic setting are limited and testing may be hindered by visual disorders and other disabilities. The functional impact of CVI/PVD can be identified by careful observation of behaviour, aided by structured history taking using open or closed questions. Positive unprompted answers tonon-leading questions (open inventory) can be helpful in eliciting characteristic visual behaviours resulting from severe visual impairment,(89, 126, 127) visual field defects, cognitive and perceptual visual disorders, and has been validated. Structured (closed set of questions) history taking using the Insight Questions Inventory (IQI), a 52 item inventory which has been used in the UK, Bangladesh and India, (68, 90, 91)and has been shown to be helpful; responses correlate with psychophysical tests of visual perception and quality of life scores. In addition, the responses lead to specific visual support strategies aimed at modifying behaviour and/or the environment to compensate for the visual problems and minimize their impact. The IQI assesses seven domains of visual function: visual field, perception of movement, visual guidance of movement, visual search, visual attention, recognition and navigation, and behaviour in crowded environments. A simple software program linksinventory response to the appropriate visual support strategies so that each child/family has a set of recommended visual support strategies

(hereafter called IQI vision support strategies) specific for that particular child. Despite anecdotal evidence that parents find the inventory andstrategies useful, there is no clinical trial evidence of the effectiveness of these strategies on improving visual function in everyday life and quality of life compared to standard ocular treatments such as the use of spectacles and strabismus surgery when required.

Although closed questions lend themselves to specific strategies, responses may be subject to

cultural bias. In this study, we will compare findings using the open and closed questions. The use of targeted, tailored strategies in the management of CVI is in line with patient centred care.(128) The parents and caregivers of the child with CP and CVI are part of decision making, and implement the intervention. Several clinicaltrials to improve the quality of life of children with CP have been undertaken but none have addressed improvement of visual function.(129, 130)

The paediatric QOL literature includes both generic and condition-specific instruments. The PedsQL 4.0 Generic Core Scales encompasses the following four areas of functioning: physical (8 items); emotional (5 items); social (5 items) and school (5 items). It was used in a similar crosssectional study in Bangladesh in which 180 children were recruited where 57 (32%) had visual acuity impairment and 95 (53%) had some parent-reported PVD. PVD was reported to be an important contributor in reducing quality of life in children with CP, independent of motor disability and impaired visual acuity.(91)

The 35-item PedsQL 3.0 CP Module encompasses seven scales: 1. daily activities (9 items); 2. school activities (4 items); 3. movement andbalance (5 items); 4. pain and hurt (4 items); 5. fatigue (4 items); 6. eating activities (5 items); and 7. speech and communication (4 items).

4.2.1 Rationale

The rationale of the study is to test the following hypotheses:

1. Structured history taking from parents or care givers (hereafter referred to as "carers") is an effective way of determining whetherchildren with CP have CVI/PVD.
2. If explained to carers and carefully applied, visual support strategies, tailored to the question inventory responses, can minimize theimpact of CVI/PVD on the child's

life and hence improve quality of life and everyday visual function.

4.3 Materials and Methods

4.3.1 Aim and objectives

The aim of this study is to improve the management and outcome of CVI/PVD in children 4 to less than 16 years of age with CP in a lower-middle income country.

4.3.2 Objectives

1. To improve the management and outcome of CVI/PVD in children 4 to less than 16 years of age with CP through the administration of theinsight questions inventory (IQI) visual support strategies.
2. To determine the effect on the quality of life and visual function of children 4 to less than 16 years of age with CP following the application ofspecifically targeted IQI visual support strategies compared to general supportive management in Cross River state Nigeria.
3. To determine whether the degree of motor or visual dysfunction is associated with the degree of improvement in quality of life.

4.3.3 Trial design

This is a parallel group, double blind clinical trial with equal arm allocation, with a superiority design. This prospective community study will beconducted in 18 local government areas in Cross River State, Nigeria. The target population are children with CP, identified in the community by key informants and confirmed by a paediatric neurologist (see below). Figure 1

shows the SPIRIT flow diagram for the effectiveness of visual support strategies for visual impairment in children with CP showing the enrolment, intervention and assessment.

4.3.4 Eligibility Criteria

Inclusion and exclusion criteria

Inclusion: Children aged 4 to <16 years diagnosed with CP (by a paediatric neurologist who would using standardized diagnostic criteria) [5] ofany type or severity and their carers.

Exclusion: Children with other causes of motor disorders, children whose carers refuse to participate and children with CP who have debilitatingillness and require immediate medical care.

4.3.5 Intervention

Intervention arm: - selected IQI vision support strategies

The intervention is application of some of the IQI visual support strategies (Appendix 2) chosen by the carers based on the “often” or “always”response to the 52 IQI questions generated by the Insight software. The social worker will explain and discuss the tailored strategies with the carer, who will be asked to select eight visual support strategies which they consider to be the most important, relevant and practical to implement. They will be asked to select 8, and 1-3 of the most important to start with, and those which could be implemented thereafter. They will be advised to practice the intervention three times a day. Carers will be given a list of all the strategies they have selected. To improve adherence and as part of the intervention, carers will be contacted by phone call every two weeks (at 2, 4 and 6 weeks). Each phone interview

will be conducted by the social worker to assess progress in implementing the strategies, to identify challenges in implementation and to remindthem of the additional strategies they selected and encourage their application and return for assessment. A check list of questions will be used.

Control arm: No IQI vision support strategies.

No intervention will be given during the 6 weeks after recruitment. After 6 week follow up assessment has been completed, carers of children inthe control arm will be offered vision support strategies based on their response to the IQI.

4.3.6 Follow-up

The trial period is 6 weeks. The flow diagram of the schedule of enrolment, intervention and assessment (Figure 1.1) and general overview of themethods (Table 2) are described.

Table 4.1 Overview of methods at different stages in the trial

Before recruitment		
Identification of children with motor impairment in the community		
Examination to confirm presence of CP		
Examination to exclude those ineligible		
Eligibility confirmed		
Recruitment with informed consent		
Ocular examination (visual function)		
Distance visual acuity (Lea symbols)		
Near vision acuity (Lea symbols)		
Visual acuity (Peek /Peekaboo)		
Grating Acuity (Lea paddles)		
Contrast sensitivity (Hiding Heidi)		
Colour vision assessment (Ishihara chart)		
Visual Fields assessment (Lea Flicker wand 280000)		
Stereopsis (Butterfly stereo acuity test)		
Extraocular motility assessment		
Vergence test		
Smooth pursuit assessment		
Saccades assessment		
Ocular alignment		
Assessment of accommodation (near pupillary reaction)		
Cycloplegic refraction		
CVI/PVD assessment (functional vision assessment)		
Visual fixation		
Visual guidance, colour, visual memory		
Visual guidance, 3D recognition of concrete objects		
Visual recognition and line orientation		
Visual recognition of size, length and direction of lines		
Visual attention		
Visual search		
Facial recognition		
Baseline interviews		
Open questions inventory		
IQI inventory (Closed)		
PedsQL 3.0 CP		
PedsQL 4.0 CP		
Strength and difficulties Questionnaire		

Data management		
Data entered into database		
Randomization	Intervention arm	Control arm
Baseline		
IQI inventory	X	X
Strategies selected by carers	X	-
Phone calls @ 2,4,6 weeks	X	-
At 6 weeks		
PedsQL 3.0 CP	X	X
Peds QL 4.0 Generic	X	X
Strength and Difficulties Questionnaire	X	X
IQI Inventory (Closed Questions Inventory)	X	X
Strategies selected by carers	-	X

Other support strategies such as wheel chairs, spectacles, and strabismus surgeries will be offered at the end of the trial to both groups of children.

Figure 4.1

	Enrolment	Randomization	Allocation	Post-allocation (weeks)					Close-out
TIMEPOINT	Pre-allocation	1 day after recruitment	3 days later	T_1	T_2	T_4	T_6	T_7	t_x
ENROLMENT									
Eligibility screen	X								
RECRUITMENT									
Informed consent	X								
Interviews [Open Question Inventory, IQI, PedsQL3.0 CP & PedsQL 4.0 Generic, SDQ]	X								
Visual function tests	X								
Randomization		X							
Allocation			X						
INTERVENTIONS									
Intervention arm: Insight vision support strategies + phone calls]				X	X	X	X		
Control arm									
OUTCOMES at 6 weeks								X	
Interviews [Open questions Inventory, IQI, PedsQL3.0 CP & Generic 4.0, SDQ]									
Both arms: Standard therapy [glasses, wheel chair, strabismus surgery]								X	
Control arm: Insight vision support strategies								X	
CLOSURE									X

SDQ = Strength and Difficulties Questionnaire

X= Means of identification of time point

Figure 4.1: The SPIRIT flow diagram the schedule of enrolment, intervention and assessment

4.3.7 Outcome measures

The primary outcome is a comparison of a change in PedsQL 3.0 CP mean score between baseline and follow up at six weeks between the twoarms of the trial. The PedsQL is being used as the primary outcome measure because there is a clear conceptualization of the health-related (HR) quality of life construct for children with CP, which reflects concepts in the assessment of PVD.

Secondary outcomes include change in functional vision, using the mean scores in the Insight Questions Inventory, the PedsQL 4.0 genericinstrument and in the strength and difficulties questionnaire between baseline and six weeks between the two arms of the trial.

4.3.8 Sample Size Calculation

Data from a pilot study in Bangladesh was used in the sample size calculation. In this study of 180 children with CP, an improvement in qualityof life, measured by PedsQL 4.0 generic of approximately 0.3SD was noted in the study population.(91) Using the Altman nomogram,(98) a sample size of approximately 370 children with CP, with 185 children in each arm, is needed to detect an effect size of 0.3SD with 80% power and at a 95% confidence, allowing for 5% loss to follow up.

4.3.9 Identification of study population and recruitment

Recruitment will take place over 12 months between 2017 and 2018. In the first stage, children with CP will be identified in the communityusing the key informant method. In the second stage, eligible children will be recruited into the trial.

Table 4.1 gives a general overview of the research stages.

Identification of children with CP

A community based sample of children with CP will be identified and recruited to reduce the selection bias inherent in health facility based studies. The Key Informant Methodology (KIM) has been shown to be effective for identifying children with severe visual impairment and blindness and children with disabilities,(35, 36) including those with CP, in lower–middle income countries. The ten questions questionnaire has been validated in an African community for use in the identification of children with neurodevelopment disorders including CP.(37) The KIM involves identifying recommended, respected community members who have had no previous medical training e.g., community leaders, teachers, local health or development workers. The key informants will undergo one day of training by the Principal Investigator using the ten question questionnaire as a screening tool.(37) After training, they will use a range of approaches to identify and list children who might have CP over a two-week period. Children they identify and their carers will be invited to a pre-agreed site, usually a primary health centre, for comprehensive history and detailed examination, including neurological examination and confirmation of the diagnosis and classification of CP by a paediatric neurologist who would use a diagnostic criteria [5].Children with CP who have debilitating illness will be referred immediately to

the tertiary health facility at the University of Calabar Teaching Hospital and not recruited. The physiotherapist will assess each child's functionsusing the GMFCS, MACS and CFCS and determine the mobility aids required. Children who need ophthalmic interventions such as corrective glasses, and strabismus surgery will be scheduled for intervention after the period of the study (It would not be feasible to provide this sooner than 6 weeks even if the children were not enrolled in the study).

Data on all eligible children i.e. name, age and gender of the child, address and phone number(s) of carers will be entered into a passwordprotected database. Each child will be allocated a unique identification number.

Informed Consent and assent

Written informed assent will be obtained from carers and consent by children who are old enough and who are able to understand what is being asked of them. Consent to participate in the interviews will also be taken from carers. Consent will be obtained after a full verbal explanation ofthe study has been given by a dedicated research officer and the key informant who identified the child, an information leaflet has been offered and time allowed for questions, (Information leaflet). The right of the child or carer to refuse to participate without giving reasons will be respected, and it will be made clear that carers and/or their child are free to withdraw from the trial at any time without giving reasons with no repercussions to them or their child. If carers withdraw before randomization, another child will be recruited. If a carer withdraws their child during the study, reasons will be sought. Before giving assent carers will be informed, that should they subsequently withdraw, all the data collected prior to their withdrawal will be maintained and used for the purposes of the study. The PI will be responsible for ensuring that all participants are protected and can participate voluntarily in an environment free from coercion or undue influence.

4.3.10 Visual function tests

The CVI and PVD examination questionnaires will be completed by the ophthalmic nurse, optometrist and the ophthalmologist. Subsections ofthe form include demography, history, visual and motility assessments, CVI/PVD examination, ophthalmological examinations, diagnosis and management plan. Table 11.2 refers to the methods of assessment of visual

function the test to be used and the person to perform the test.

The following criteria for prescribing glasses will be used: myopia ≥ -2.0D, hypermetropia ≥ +4 D and astigmatic cylinder ≥ -2.0D. Anterior segment examination will be done with a portable slit lamp (Reichert Technology) and dilated posterior segment examination will be performedby an ophthalmologist using a binocular indirect ophthalmoscope (Appassamy) with a 28D or 20D lens. Fundus photographs will be taken usinga Horous fundus camera. Visual fields will be assessed using the Lea wand.

Table 4.2: Visual functions and methods of assessment

Visual function	Test	Performed by
Presenting distance visual acuity	Lea symbols	Optometrist
Presenting distance visual acuity	Peek visual acuity test; Peekaboo visual acuity test	Optometrist
Presenting near visual acuity	Lea symbols	Optometrist
Grating acuity	Lea paddles	Optometrist
Contrast sensitivity	Hiding Heidi	Optometrist
Colour vision	Acquired and congenital colour chart	Optometrist
Visual fields	Lea Flicker Wand 280000	PI
Stereopsis	Butterfly stereo acuity test with lea symbols	Optometrist
Eye movements		PI
Vergence	Observation of binocular movements of the eye i.e., convergence and divergence, using a fixation target.	PI
Motion processing-smooth pursuit (horizontal and vertical)	Uncooperative or inattentive children are tested by slowly rotating a mirror that measures 10cm by 15cm held before their eyes. Verbal children: the patient is asked to track the tip of a blue ball point pen held a meter before the eyes with the head still. The target is moved at a low, uniform speed.	PI
Motion processing-saccades	Non-verbal: The infant or child is enticed to look at the tester's face. When the fixation is in the midline, one of the objects is presented at about 20-30cm from the midline. The infant or child is again enticed to look at the tester's face after which the other object is presented on the other side. Verbal -The patient is asked to look at the examiner's nose and then at the examiner's finger to the left or right of central fixation only upon verbal command.	PI
Strabismus	Corneal reflex test and Modified Krimsky	PI
Accommodation	Near Pupillary Reaction Method	PI
Cycloplegic Refraction	Cyclopentolate 2% + Phenylephrine 2.5% Zeiss autorefractor/keratometer Manual objective retinoscopy	Optometrist PI

4.3.11 Assessment of CVI/PVD

CVI examination will be conducted using several tests. [40,41, 42, 43], with observation of the child's performance (Appendix 4 and 5). Table 3describes the tests used to assess CVI/PVD. Each observation has a score based on the ability or inability of a child to perform, and all will be undertaken by the researcher (RD). After examinations, participants and their care givers will disperse to their homes and await further contact by phone call.

Table 4.3: Assessment for PVD, corresponding test, observations and grading by PI

Description of CVI/PVD	Test	Observation	Grading
Visual fixation	A fixation light is presented before both eyes at a distance of 0.75m	Central, sustained or maintained	Present (central, sustained & maintained) or absent
Visual guidance, colour, eye hand coordination and visual memory	Lea 3-D puzzle coloured	Eye-hand coordination and visual guidance of movement: watching the movement a child makes with their hand, elbow or forearm when asked to turn over a piece of puzzle. The ability to place the puzzle pieces correctly. The capacity to orientate the shape is subjectively viewed. Short-term memory for localisation: can a child place a piece of puzzle in the correct place after being distracted.	Easy, difficult, cannot do and why, or not applicable
Visual guidance and 3 D recognition of concrete objects	Lea 3-D puzzle black & white	Eye-hand coordination and visual guidance of movement: watching the movement a child makes with their hand, elbow or forearm when asked to turn over a piece of puzzle. ~3-dimensional assessment of an object. Ability to place the puzzle pieces correctly. Short-term memory for localisation: can a child place a piece of puzzle in the correct place after being distracted	Easy, difficult, cannot do and why, or not applicable
Visual recognition and line orientation in three dimensions (vertical, horizontal, oblique) & eye hand coordination	Lea mailbox game	Visual perception of line orientation done with the fingers, hand, or forearm: • the child is asked to drop a card through the slot of the LEA Mailbox Game • the child is asked to match the orientation of the slot	Easy, difficult, cannot do and why, or not applicable
Visual recognition of differences in size, length and direction of lines	Lea rectangles game	Size, length perception: • Watch the orientation of the hand and fingers as the child grasps a block • can the child place a block of the same length on top of another?	Easy, difficult, cannot do and why, or not applicable

		• Can the child appreciate short and long as a change in length? • Response to the enquiry concerning whether a correct or an incorrect arrangement looks the same	Yes, No, not sure, do not know, not applicable
Visual attention	Identification of self reflection in the mirror test	Identification of self-image The distance at which the child loses interest in self image.	Yes or No Documentation of the distance at which. Attention is lost.
	Pick-up test: Picks up a small ball of uniform size (1mm) placed on a patterned surface	Searches for and identifies the ball.	Easy, difficult, cannot do and why, or not applicable
Visual search	Pick-up test: Picks up a small ball of uniform size (1mm) placed on a plain surface	Searches for and identifies the ball.	Easy, difficult, cannot do and why, or not applicable
Facial recognition Test	Heide expression test	Interpretation of facial expressions: Can the child interpret the facial expression? Can the child identify a similar facial expression?	 Yes, No, cannot do, why or not applicable Easy, difficult, cannot do and why, or not applicable

4.3.12 Randomization

Baseline examinations are performed before randomization so that stratification and blocking can occur and also, so that those collecting thebaseline data are masked to the allocation.

Sequence generation

The database of children recruited is accessible to the researchers and contains information on their unique ID, age, sex and GMFC score. The database will be sent to the data analyst in the University of Calabar Teaching Community Medicine Department at the end of the examination per local government area consecutively. Children will be stratified and blocked by age group 4-9 and 10-<16 and by Gross Motor Function Classification Score (GMFCS) Level 1-3 and 4-5. The randomisation sequence will then be generated by the data analyst (SA) using Stata 11 programming syntax for block randomisation of patients into the treatment and control group. The randomization database will be password protected, and only accessible to the data analyst. The randomisation database holds data on patients' unique ID, block number, block sizes andtreatment vs. control groups. The data analyst will merge the randomisation database with the patient database using the patients' unique ID which will be the same in both datasets. The result of randomisation in the combined dataset will be sent to the research officer for the purposesof patient enrolment into the randomised clinical trial (RCT).

Allocation concealment and implementation

Carers of children assigned to the treatment arm of the trial will be contacted by phone call and personal contact by the key informant within 3 days of recruitment by an independent research

clerk who is masked to the baseline examination. Carers will be invited to visit the same primary health centre where the baseline examination was conducted at an agreed time convenient to the carers. At this visit the intervention willbe selected by the carers and explained to them by the social workers.

4.3.13 Contamination and masking

This is a double blind study. It is unlikely that contamination will occur, as allocation is not being done on the same day as the examination, and carers of children in intervention and control arms are unlikely to meet after the baseline assessment. The research assistant who is not part of theexaminations will place calls to care givers within 3 days of the examination. The data analyst involved in the sequence generation is not involved in any of the field work. No member of the team collecting baseline data will be involved in explaining the intervention nor in collecting outcome data. part of the treatment teams or final evaluation team. Parent or care givers who do not respond to phone calls will be personally contacted by the key informant. The PI will not know the arm to which children are allocated to. The randomisation sequence and

allocation is computer generated and cannot be changed. The 6th week outcome interviews will be performed by a different set of researchersfrom those who collected pre-intervention data. This follow up assessment will be a mix of children in the intervention arm and in the control arm attending on the same day.

4.3.14 Data collection methods

Interviewers will be trained for 6 weeks on data collection using the following study instruments: PedsQL3.0 CP, PedsQL4.0 generic, SDQ, IQIand strategies, strategy follow up form.

Data will be collected at base line and at the end of follow up after 6 weeks. Outcome data will be entered into one Microsoft excel database by amasked medical records officer who has no knowledge of, or access to, identifiable participant information or treatment assignment.

Open ended and Insight Question Inventories

All carers of children with CP, will be interviewed with both the open ended questions inventories (Appendix 6) first and the IQI second at baseline and at the end of six weeks. The modified Insight inventory, a 52-item questionnaire will be administered to each carer giver. There are 7 sections. Responses to each question will accord with a 5-point Likert scale to describe whether a child has problems: never, rarely, sometimes,

often or always including “not applicable”. These responses will be awarded numbers from 1 to 5 respectively. The questions in each section aredesigned to identify CVI/PVD. Visual tasks involve both the dorsal and ventral visual streams functioning together, however, the activities askedabout in sections1, 2, 3, 4 and 5 are designed to be illustrative of mainly dorsal visual stream abilities and whilst section 6 and 7 relies more on ventral stream function.

Quality of Life– methodology

For the PedsQL 4.0 generic tool and the PedsQL CP 3.0 module, the parent’s proxy form will be used.

The 23-item PedsQL 4.0 Generic Core Scales encompass: (1) Physical Functioning (8 items); (2) Emotional Functioning (5 items); (3) Social Functioning (5 items); and (4) School Functioning. While the 35-item PedsQL 3.0 CP Module is composed of seven scales, namely: (1) Daily Activities (9 items); (2) School Activities (4 items); (3) Movement and Balance (5 items); (4) Pain and Hurt (4 items); (5) Fatigue (4 items); (6) Eating Activities (5 items); and (7) Speech and

Communication (4 items). The parents will be given a Likert response scale of 1) Never 2) almost never 3) sometimes 4) often 5) almost always to choose one single option. The results will be entered on the PedQL form.

The results from the questionnaire will be transferred to an excel sheet to number 0 to 4 respectively. To interpret the scores, items are reversed scored and linearly transformed to a 0-100 scale, so that higher scores indicate better HRQOL (Health-Related Quality of Life). To reverse the score, the 0-4 scale items will be transformed to 0-100 as follows: 0=100, 1=75, 2=50, 3=25, 4=0. To create the scale scores, the mean will be computed as the sum of the items over the number of items answered (this accounts for missing data). The number of missing values in the scale(call it miss) will be summed and the item scores divided by the number of items in the scale minus those that were missed. The Psychosocial Health Summary Score, mean will be computed as the sum of the items over the number of items answered in the Emotional, Social, and School Functioning Scales. The total scale score, for both the generic and CP modules, will be obtained by computing the mean as the sum of all the items over the number of items answered on all the Scales.(131) If more than 50% of the items in the scale are missing, the Scale Scores will notbe computed. If 50% or more items are completed they will be used to impute the mean of the completed items in a scale according to PedsQL guidelines.

Strength and difficulties questionnaire

The strength and difficulties questionnaire interview will be conducted after the PedsQL CP module administered at the examination site. It has 25 items on psychological attributes which are divided between 5 scales: emotional symptoms, conduct problems, hyperactivity/inattention, peerrelationship, pro-social behaviour. The parents/caregivers will be given instructions to give their answers on the basis of the child's behaviour over the last six months.

4.3.15 Data management

The data monitoring committee, which is independent of the sponsors, will provide oversight of the data collection process and will meet after recruitment has been completed in each local government area. Two optometrists and two ophthalmologists will undergo competency training inclinical examinations to ensure standardization of data collection methods and completing the ophthalmic examination questionnaire. Two nurses will be trained on history taking using the open questions inventory, questionnaires including, PedsQL Generic and CP Module and Strength and difficulties questionnaires. Four social workers will be trained to administer the IQI and strategies and follow up form. Two nurses will be trained to conduct the sixth week follow up assessment. Successful completion of competency training for all research staff will be confirmed by a competency checklist.

Participant data will be stored securely and their confidentiality protected. Each participant will be allocated a unique trial number at the end ofexamination and this will be used throughout the study to identify all data relating to the participant. Data collected by all members of the research team will be entered on paper then transferred to a Microsoft Excel data sheet on a protection compliant drive in the research office.

Questionnaires and inventory questions will be assessed at the end of the research day. Where there are missing data, every attempt will be madeto complete the data. Data will be entered the following morning. Data entry errors will be corrected at the end of each following day. Data will be updated to the Chief investigator every day data are collected. All patient identifiable information will be kept on encrypted computers in a secure office, with access limited to authorised members of the research team. All data will be archived in secure archive facilities in the London School of Hygiene & Tropical Medicine for ten years. After this time, all documentation will be kept in a general data pool in LSHTM. The finaldataset will initially be available only to the study team. After the results of the trials have been reported, the trial data will be made publicly available through the LSHTM repository.

4.3.16 Statistical methods

Missing data will be addressed at the analysis stage. Statistical analyses will be conducted with STATA (version 11.0) as an intention to treat analysis. Our descriptive statistics will include means and SDs, medians and interquartile ranges for continuous variables, and the number andproportions for categorical variables as appropriate.

We will compare the two intervention groups at baseline regarding characteristics and demographics, using two-tailed Student's t tests and theeffect size to test the strength of the difference. Variables that differ between the groups at baseline will be considered as possible confoundersand adjusted for in subsequent analyses. The alpha level will be set at 0.05 for all comparisons.

We will use the unpaired (two- sample) t-test- for comparing the change in PedsQL score 4.0 and 3.0 CP module score and IQI score in arm 1 versus arm 2. Skewed data will be analysed with nonparametric test alternatives (e.g. we will use the Mann-Whitney U test instead of

Student's tests if data are skewed).

2. Paired t test for comparing mean scores of PedsQL CP 4.0 before and after intervention.

3. Paired t test for comparing mean IQI scores before and after intervention.

4. Potential effectors of PedsQL 3.0/4.0 and IQI (visual function) outcome, such as, socio demographics; all medical and clinical variables such as; GMFCS, MACS, CFCS, grade of vision, key CVI/PVD examinations and types of CP will all be investigated pairwise and those found to becorrelated will be entered into a multivariate regression model.

5. Subgroup analysis – which type of children with CP and what level of GMFCS showed improved quality of life – analysis by socio- demographic factors, age, sex, type of CP, GMFS, MACS, CFCS, VA, type of PVD by insight domain and association with ocular examinations.

4.3.17 Monitoring

Harms

Reported adverse events such as pain or distress will be carefully monitored throughout the trial, although minimal risk is anticipated. Contactsfor reports of adverse reactions will be put in place and carers advised to report to the study centre- The Calabar Childrens' Eye Center. Reporting of the adverse events will follow LSHTM guidelines on recording, managing and reporting adverse events for behaviouralinterventions.

Loss to follow up

Participants who do not respond to the two weekly phone calls by the sixth week will have a home visit by the key informant and the end of trialquestionnaires will be administered at their home by the research team members. Participants who are lost to follow up will be included in the analysis.

Audit

Data will be reviewed at the end of the field work daily. Random comparisons between entered data and filled protocols will be performed after each 5 entries. This will be done independently of the sponsors. The study may be subject to audit by the London School of Hygiene & Tropical Medicine under their remit as sponsor, the Study Coordination Centre and other regulatory bodies to ensure adherence to Good Clinical Practice.

4.4 Discussion

The EVSSCP trial study will be the first RCT to investigate the effect of tailored visual support strategies for the treatment of perceptual visualdisorders in children with CP. Evidence based management strategies for children with this problem are urgently needed.
Considering the lack of awareness of visual support strategies in the treatment of perceptual visual disorders and the need for knowledgedissemination in this area, this trial should lead to a significant advance in the management of this condition.
All children with CP are being recruited to the trial not just those with confirmed CVI. The reason for this is that subtle visual problems are common in CP and since detailed visual assessment is not always possible in practice it would be useful to have a tool that can be given to all CPfamilies. The tool is self-titrating in that if no visual problems are reported in the IQI then no strategies will be given. This could potentially reduce the power of the trial but if the tool is found to be useful for all children with CP then it will be clinically more useful.
Although pilot data from Bangladesh has been encouraging, there have been no RCT that measure the effects of potentially effective visual support strategies in habilitating children with

PVD. If it were shown to be effective it could be scaled up as a community based intervention. Inthis study there are limitations in the use of some examination methods.(132)

Limitations

The EVSSCP trial is planned to be carried out as a single country study and can, therefore only be seen as a preliminary study in Africa.Trial status
The protocol version number 2 of 18/09/2016 recruitment began on the 4th of December 2017. Recruitment is expected to be completed by theend July 2018.

List of abbreviations

CRS: Cross River State; LGA: Local government area; CP:Cerebral Palsy; CVI: Cerebral visual Impairment; QoL:Quality of Life; IQI: Insightquestions inventory; PVD: Perceptual Visual Disorder/Dysfunction; PedsQL: Paediatric Quality of Life; EVSSPVD: Effectiveness of Visual Support Strategy for perceptual visual dysfunction; KIM: Key informants methodology; GMFCS: Gross motor function classification scale; MACS: Manual ability classification scale; TQQ: Ten questions questionnaire; RCT: Randomized control trial; SDQ: Strength and DifficultiesQuestionnaire VA: Visual acuity

Declarations

Research ethics approval

The study will be conducted in accordance with the Helsinki Declaration. Approval has been received from the Cross River State Ethics committee of the Ministry of Health (CRS/MH/HREC/015/Vol.V/211) and from the LSHTM Research Ethics Committee (LSHTM Ethics Ref:11678). Pan African Trials registry number is (PACTR201612001886396)
Submission of protocol, progress reports and notification of end of study will be done using the

SPIRIT guideline [45]. This protocol also follows the SIRIT checklist (figure 2) and CONSORT statement.

Important protocol modifications will be communicated to the investigators, REC/IRBs, trial participants, trial registries and journals.Publication policy

Authorship will follow the recommendations of the International Committee of Journal Editors for authorship. All publications and presentationsrelating to the study will be authorised by the Chief Investigator. The first publication of the trial results will be in the name of the Research team. If there are named authors, these will include at least the trial's Chief Investigator and principal Investigator. We plan to disseminate the results of this study to the scientific, medical and general public by publication in national and international peer-reviewed journals, as well as bypresentations at conferences and meetings. The list of potential articles includes: Trial methodology (Trials), outcome of the trial and baseline description of QoL in children with CP in Nigeria and its predictors, literature review of CVI/PVD in children with CP in Nigeria and baseline description of visual and ocular morbidity in children with CP in Nigeria and its predictors.

Confidentiality

Any participants' identifiable data collected by the Study Coordination Centre will be stored securely and their confidentiality protected inaccordance with the Data Protection Act 1998. Participants record on the data sheet will be by represented numerically.

Indemnity

London School of Hygiene & Tropical Medicine holds Public Liability ("negligent harm") and Clinical Trial ("non-negligent harm") insurancepolicies which apply to this trial.

Sponsor

London School of Hygiene & Tropical Medicine will act as the main sponsor for this study. Delegated responsibilities will be assigned locallyand will not authority over the study management, analysis and publication

Funding

The Queen Elizabeth Diamond Jubilee Trust/Commonwealth Eye Health Consortium. London School of Hygiene and Tropical Medicine GrantCode: ITCRZC6814. Treatments will be funded by the University of Calabar Teaching Hospital, Calabar. Funding:

Trial management

The day-to-day management of the trial will be coordinated through the Study Coordination Centre.

Trial closure

The EVSSCP trial should take a period of one year. Trial closure is anticipated in the last quarter of 2018 after the following have been done.

1. After the administration of the appropriate Insight strategies to the families in arm 2 at the end of six weeks.

1. After all questions posed by participants have been answered.

2. After all benefits have been given to each child.

3. After outcome data from the last child recruited have been obtained

Availability of data and materials

Public access to the protocol will be provided via open access of the article. The dataset

supporting the conclusions of this article will beavailable upon completion of the study and on reasonable request to the corresponding author.

Authors' contributions RB planned the study and developed the protocol, drafted the manuscript and checked the final draft of the manuscript.RD planned the study and developed the protocol, drafted the manuscript and checked the final draft of the manuscript. KB contributed to developing the protocol, drafted the manuscript and checked the final draft of the manuscript. DM contributed to the methods and statistical analysis plan. CG contributed to developing the protocol, drafted the manuscript and checked the final draft of the manuscript. GD contributed to developing the protocol, drafted the manuscript and checked the final draft of the manuscript. All authors read and approved the finalmanuscript.

Acknowledgments

We acknowledge all the families with children who had CP and the key informants who participated in this study.

Competing interests: The authors declare that they have no competing interests

3 Cultural and language adaption of the Insight History Question Inventory and Visual Support Strategy for use in the Cross River State, Nigerian population: a pilot study

Authors

Roseline E Duke*1,2 Bassey Okon,3 Esther Archibong,4 Richard Bowman 1

Affiliation

1London School of Hygiene and Tropical Medicine, International Centre for Eye Health,

2 University of Calabar Teaching Hospital, Children's Eye Centre, Department of Ophthalmology, Calabar, Cross River State, Nigeria.

3 University of Calabar Department of Linguistics, Calabar

4 University of Calabar Department of Sociology, Calabar

* Corresponding author: Roseline E Duke roselineduke@unical.ac.ng
Calabar Childrens Eye Centre, Department of Ophthalmology University of Calabar Teaching Hospital, Calabar
Cross River State, Nigeria

Keywords

cerebral palsy, perceptual visual dysfunction, cerebral visual impairment, Insight Questions inventory, vision support strategies, Nigeria

1.2 Chapter Abstract

Cerebral visual impairment (CVI), including perceptual visual dysfunction (PVD) is common in children with cerebral palsy (CP). Identificationof CVI including PVD is most often times difficult to make. The Insight Question Inventory (IQI), a clinical questions inventory is employed clinically for eliciting visual behavioural problems in children CVI/PVD. It is accompanied by matched visual support strategies (IQI VSS) as anintervention. The IQI has been used in several countries including Britain, India and Bangladesh. However, there is no experience of using the IQI/VSS in sub-Saharan Africa (SSA). The study sought to translate/adapt the IQI and report the suitability for use in Nigeria.

A mixed method study was conducted among carers of children with CP. Cultural and linguistic modification of the IQI was undertaken throughsemi-structured questioning, observation of child's behaviour at home and clinical use of the inventory.

We found that the IQI can be adapted for use in this population. Parental education to characterise CVI/PVD in this population is needed. The implications for the use of the IQI would involve a detailed health education programme for parents and carers on the symptoms and reporting ofCVI/PVD prior to the application and use of the questions inventory in a population of parents were CVI/PVD is suspected.

1.3 Introduction

The Insight questions inventory was approached initially as a questionnaire. Beyond the use of the questions as a questionnaire, is its use asquestions inventory to identify, quantity, describe, proffer solution to PVD dysfunction and measure the impact in the life of the child.

Following this development, the next priority internationally would be to agree to the questions and strategies in order for the IQI and visual support strategies to be used in international clinics, as well as a valid research instruments in international clinical trials. For this to happen, itmust be culturally adapted and the translations need to be linguistically validated for the targeted population.

The validation process should produce the outcomes of a language and culturally adapted Insight Questions Inventory (IQI) to a Nigeria version,parental awareness of PVD, parental understanding of the IQI, parent's ability to remember the strategies and should be able to be used to assess outcome of visual function and quality of life after the use of the visual support strategies.

We report the development of a culturally and linguistically adapted IQI and its implications for use in children with CP and CVI/PVD inNigeria.

1.4 Materials and methods

A mixed method pilot study was conducted in Calabar municipality in Cross River State, Nigeria in between November 2015 to June 2016. Qualitative investigations were conducted using purposive sampling. Triangulation was achieved using the following methods: semi-structuredquestions and observations.

1.4.1 Study population

Parents of children with cerebral palsy and the children from the Calabar municipality community were identified and invited to the CalabarChildrens Eye Center, University of Calabar Teaching Hospital for interviews and their children for ocular examinations.

1.4.2 Sample size/recruitment of patients

The sample size for the quantitative aspects of the cross sectional field study was calculated using Leslie Kish sampling technique for a population >10,000.00 (Calabar municipality is about 300,000)(133) expected frequency of 0.5% for CP, degree of precision of 0.05, standard error of 1.96 and a non-response rate of 10% gave a population sample size of 24. Recruitment of parents and their children with CP was done

using the Key Informant method. Following the presentation of the children found in the community by the Key Informants to the Calabar Childrens Centre of the University of Calabar Teaching Hospital, the pediatric neurologist examined all the children to ascertain those who hadCP. Children with a confirmed diagnosis were included into the study. The parents participated in the translation/adaptation process and the children had ocular examinations conducted.

Instrument development

Developing a culturally and linguistically appropriate version of the Insight Question Inventory involved several stages.

1.4.3 Linguistic translation and cultural adaptation

The first stage was to perform the linguistic translation and cultural modification of the English version of the IQI and strategies (Appendix 1 and 2) using the International Test Commission Guidelines on Translating and Adapting Tests for the translation/adaptation of existing tests and

instruments (110, 134). The following steps were undertaken: translation (produced IQI version 1), forward translation (produced IQI version 2),

reconciliation, backward translation (produced IQI version 3) and review of back translation, harmonization, clinic testing (produced IQI version4) through cognitive debriefing, field study for further cognitive debriefing and proof reading, resulting in the production of the final modified version of the Nigerian IQI/VSS (IQI version 5).

Translation

The aim of the linguistic validation and cultural adaptation was to produce a version in the 'Nigerian English language and expression' (our target language). Translations were done by a linguistics expert and a social scientist. Each translator independently produced a Nigerian versionof the IQI and strategies. A face validation and comparison of both translated IQI was performed by a reconciliation committee, comprising the chief and principal researchers, the research coordinator and research assistant. After discussion of each item, a final version of each question was selected. (IQI version 2).

Backward translation

The backward translation and review which was done by a separate language specialist (SE), where IQI version 2 was compared to the originalIQI and strategies and harmonized with the original English version. This gave rise to the backwards translation IQI version 3.

IQI version 3 was used as an inventory to identify CVI/PVD and converted into a semi-structured questionnaire as a parent reported questionnaire to determine whether the questions were understood and should be left in its original form, modified in language or changed

entirely. Children and parents with CP were recruited from the neurology clinic of the Department of Paediatrics of the University of Calabar Teaching Hospital. To determine parents' understanding of the IQI, they were interviewed question by question and their responses recorded under one of four categories:(P1); no difficulty understanding or remembering the question; (P2) a difficulty remembering the question; (P3) adifferent understanding of what the question referred to; (P4) the respondent had difficulty recalling, formulating or reporting answer. Parents were asked to respond by thinking aloud as they decided what the question meant, what they understood and their decision (135-137). Interviews were undertaken for the 52 questions. Questions were modified or left in their original form in accordance with their responses. Thisproduced IQI version 4.

Validation of the modified IQI instrument

The IQI version 4 and strategies were field tested and piloted. Children with CP, 4-15 years in the community were identified by key informant'smethod and brought to the Calabar Children's Eye Centre of the Department of Ophthalmology, University of Calabar Teaching Hospital for ocular/visual and general examination. The IQI was administered as a parent reported questions inventory to elicit and identify CVI/PVD and a semi structured questionnaire to determine the understanding of the questions in the IQI and to determine what modifications were required in

for the adaptation of the clinical inventory. The IQI was administered by the principal investigator. Data analysis from the pilot study resulted in the final Nigerian version to be used in a larger clinical trial. Children identified in the community with other causes of motor impairment were excluded from the study by the Paediatric Neurologist. Data on clinical and sociodemographic history, general and ocular examinations for cerebral palsy and CVI/PVD were collected and interviews with the PedsQl 4.0 generic and 3.0 CP were conducted . Parents

were asked to comeback after two weeks for another interview, when the IQI and the Peds 4.0 generic and PedsQL 3.0 CP would be repeated.

1.4.4 Observation

Three randomly selected homes of parents that participated in the field test were visited to observe the behaviour of children and their parents athome two weeks after the field examinations, to compare with the reports of what the parents had given. All three observers used the IQI questions to ascertain specific observations based on the IQI and also made observations notes about each child. RD (principal researcher) and two KIs made the observations between 4 -7pm daily for three days in each household. The observers were neutral.

1.4.5 IQI/Visual support strategies

Parents were encouraged to choose at least 3 tailored IQI/VSS strategies they felt would improve the well-being of their child. The strategies

were explained by the ophthalmologist and a list of strategies given to parents to administer.

1.4.6 Pediatric Quality of Life Inventory(PedsQL)

The PedsQL is a modular instrument for measuring health-related quality of life (HRQOL) in children and adolescents ages 2 to 18 years and has been used in randomized clinical trials successfully. The PedsQL inventory (PedsQL) 3.0 CP module has 35-item which encompasses sevenscales: (1) Daily Activities (9 items); (2) School Activities (4 items); (3) Movement and Balance (5 items); (4) Pain and Hurt (4 items); (5) Fatigue (4 items); (6) Eating Activities (5 items); and (7) Speech and Communication (4 items). The module is reported to be reliable,

valid and sensitive to PedsQL in pediatric CP. The parent proxy forms will be used at baseline and two weeks after. This questionnaire also describes areasof assessment in the IQI inventory.

1.4.7 Peds QL4.0 Generic Core scales

The 23-item PedsQL 4.0 Generic Core Scales encompass: (1) Physical Functioning (8 items); (2) Emotional Functioning (5 items); (3) Social Functioning (5 items); and (4) School Functioning. Giving information on the Psychosocial Health summary score. This was used at baselineand 2 weeks after.

1.4.8 Peds QL3.0 Cerebral Palsy (CP) scales

The 35-item PedsQL 3.0 CP Scales encompass: (1) Daily activities; (2) Movement and Balance; (3) Pain; (4) Fatigue; (5) Eat activities; (6)Speech and communication.
Both used the parent's proxy forms was used. This was used at baseline and 2 weeks after. The parental interview was administered by trainedresearch assistants. The Peds QL generic and CP modules were administered during the examination and two weeks after the administration of strategies by the ophthalmologist and the results compared.

1.4.9 Data Analysis

Quantitative data from the IQI analysis and the ocular examinations were analysed using descriptive statistics (frequencies, proportions, means and standard deviations) for the analysis of the semi-structured questionnaire (IQI). Each item question on the IQI is scored as described in the introduction. The mean of the scale is calculated. The mean for each child and the subsection of the questionnaire (visual field (VF), perception of movement (PM), visual guidance of movement (VG), visual search (VS), visual attention (VA), recognition and navigation (RN) for each child was calculated, a section and total mean score of ≥3 was

indicative of CVI/PVD. Data obtained from observation and was also analysed asabove to determine the mean values.

Written informed assent was obtained for each child. Ethical approval was received from the Cross River State Ethics committee of the Ministryof Health (CRS/MH/HREC/015/Vol.V/211) and from the London School of Hygiene and Tropical Medicine Research Ethics Committee (LSHTM Ethics Ref: 11678).

1.5 Results

1.5.1 Translation

The first outcome from the original Insight Questions Inventory (English and expressions) version (supplementary material 1) of the IQI is version 1. These had contextual, cultural preference and the 'local language' modifications in 32(61.5%) out of the 52 questions. Table 2. Thequestions they both were in agreement to modify were 13(25%): Questions-1,3,4,15,18,19,24,30,40,45,48,51 of the English version.

1.5.2 IQI Version 2

The translation committee reverted back to the original IQI English version 1 for question 30, as the meaning was altered by the translation.

1.5.3 IQI version 3 (Backward translation)

There were no changes made and no difference in meaning between the English version and the Nigerian version in process.

1.5.4 IQI version 4 (clinic testing)

Eight carers who had children with CP participated. There were no changes made to the IQI version 3 because no problems were identified withparental understanding of their meaning. The unchanged version is renamed IQI version 4 and was used both as a history inventory and semi structured questionnaire in the field test which involved the KIs.

1.5.5 Field test was performed where parents were recruited from the community rather than the clinic to produce IQI version 4.

Sociodemographic details are presented in Table 5.1 for carers that participated in the IQI (semi-structured questionnaire) interview. There were 23 carers of children with CP from the community. One child was unable to participate in the ocular examination due to time constraints of the parents. The semi structured interview was performed by all 23 parents who, gave responses to the IQI for all 52 questions.

Table 5.1: Socio-demography of parents who participated in the in-depth interviews

Variable	Frequency	Percentage (%)
Age group/years		
<32	11	61.1
≥33	7	38.9
Mean age ± SD	32.7 ± 5.2	
Sex		
Male	2	11.1
Female	16	88.9

Occupation of parents		
Teacher	4	22.2
Administrative staff	7	38.9
Cleaner	1	5.6
Messenger	1	5.6
Ward orderly	3	16.7
Nurse	1	5.6
Trader	1	5.6

Table 5.2 shows the proportion of parents and their levels of understanding of each of the IQI questions. The parents understanding was lowest for questions 3, 5,15, 22,33, 34 while, question 3 was forgotten by 5(22.7%) parents. Over 50% of parents in all subcategories in the IQI had nodifficulty understanding the meaning of the question or the meaning of particular words or the concepts projected (P1).

Table 5.2 Proportion of parents whose response to each IQI questions where either P1, P2, P3 or P4

Question	P1(%)	P2(%)	P3(%)	P4(%)
1	10(55.6)	2(11.1)	2(11.1)	4(22.2)
2	11(61.1)	5(27.)	-	2(11.1)
3	9(50.0)	4(22.2)	1(5.6)	4(22.2)
4	11(61.1)	2(11.1)	5(27.8)	-
5	5(27.8)	5(27.8)	5(27.8)	3(16.7)
6	10(55.6)	7(38.9)	-	1(5.6)
7	10(55.6)	7(38.9)	-	2(11.1)
8	13(72.3)	-	3(16.7)	2(11.1)
9	8(44.5)	5(27.8)	2(11.1)	3(16.7)

10	11(61.1)	1(5.6)	4(22.2)	2(11.1)
11	14(77.8)	2(11.1)	-	2(11.1)
12	12(66.7)	-	3(16.7)	3(16.7)
13	16(88.9)	-	1(5.6)	1(5.6)
14	8(44.5)	5(27.8)	5(27.8)	-
15	9(50.0)	5(27.8)	2(11.1)	2(11.1)
16	10(55.6)	4(22.2)	1(5.6)	3(16.7)
17	14(77.8)	3(16.7)	-	1(5.6)
18	14(77.8)	-	2(11.1)	2(11.1)
19	11(61.1)	5(27.8)	1(5.6)	1(5.6)
20	14(77.8)	4(22.2)	-	-
21	14(77.8)	2(11.1)	2(11.1)	-
22	1(5.6)	8(44.4)	6(33.3)	3(16.7)
23	3(16.7)	8(44.4)	3(16.7)	4(22.2)
24	3(72.2)	4(22.2)	1(5.6)	-
25	17(94.4)	-	1(5.6)	-
26	10(55.6)	4(22.2)	3(16.7)	1(5.6)
27	14(77.8)	2(11.1)	2(11.1)	-
28	16(88.9)	2(11.1)	-	-
29	14(77.8)	2(11.1)	2(11.1)	-
30	5(27.8)	6(33.3)	3(16.7)	4(22.2)
31	12(66.7)	4(22.2)	1(5.6)	1(5.6)
32	8(44.4)	7(38.9)	3(16.7)	-
33	3(16.7)	8(44.4)	5(27.8)	2(11.1)
34	8(44.4)	7(38.9)	2(11.1)	1(5.6)

35	15(83.3)	2(11.1)	-	1(5.6)
36	8(44.4)	3(16.7)	2(11.1)	5(27.8)
37	15(83.3)	3(16.7	-	-
38	13(72.2)	2(11.1)	2(11.1)	1(5.6)
39	5(27.8)	7(38.9)	4(22.2)	2(11.1)
40	6(33.3)	8(44.4)	1(5.6)	3(16.7)
41	14(77.8)	4(22.2)	-	-
42	15(83.3)	1(5.6)	2(11.1)	-
43	17(94.4)	-	-	1(5.6)
44	13(72.3)	2(11.1)	1(5.6)	2(11.1)
45	15(83.4)	3(16.7)	-	-
46	14(77.8)	4(22.3)	-	-
47	6(33.4)	7(38.9)	2(11.1)	3(16.7)
48	8(33.4)	5(27.8)	-	5(27.8)
49	10(55.6)	3(16.7)	2(11.1)	3(16.7)
50	18(100.0)	-	-	-
51	10(55.6)	2(11.1)	3(16.7)	3(16.7)
52	18(100.0)	-	-	-

P1: the respondent had no difficulty understanding the meaning of the question or the meaning of particular words or concepts P2: the respondent had difficulty remembering the question

P3: the respondent had a different understanding of what the question P4: the respondent had difficulty recalling, formulating or reporting answer

The proportions of the various responses from cognitive debriefing of the IQI inventory

subcategories are reported in details in Table 5.3: Visualfield (VF)=112/198 (55%), Perception of Movement(PM)=55/90(61%), Visual Guidance) VG) =71/126(56%), Visual Search(VS)=125/216(58%), Visual Attention(VA)=47/90(52%), Recognition and Navigation(RN)=150/198(76%) and Behaviour crowded spaces(BCS)= 64/90(71%).

Table 5.3: Description and proportion of comments made by parents for each IQI question per subcategory ofIQI

Comments	Visual Field (Q 1-11)	Perceptionof Movement (Q 12-16)	Visual guidance of Movement (Q 17-23)	Visual Search (Q 24-35)	Visual Attention (Q 36-40)	Behaviour in crowded scenes (Q 48-52)	Recognitionand Navigation (Q 41-52) (minus 48)
Total no of observations in IQI inventory	198	90	126	216	90	90	198
	N (%)	N (%)	N (%)	N (%)	N (%)	N (%)	N (%)
P1	112 (57)	55(61)	71(56)	125(58)	47(52)	64(71)	150(76)
P2	40(20)	14(16)	30(24)	48(22)	23(26)	10(11)	26(29)
P3	22(11)	12(13)	14(11)	23(11)	9(10)	5(6)	10(5)

P4	25(13)	9(10)	11(9)	10(5)	11(12)	11(12)	12(6)

Questions that respondents had difficulty recalling, formulating or reporting answer were mostly in the VF 25/198(13%) category.

The number and proportion of parents who wanted each IQI question modified, changed entirely or left in the original form. Questions left intheir original form were: 7(39%) questions (13,25,35,38,41,42,52).

Table 5.4: The number and proportion of parents that recommended each question to be left Original (O), Modified (M) or Change © N=23

Question	O (%)	M (%)	C (%)
1	16(88.9)	-	2(11.6)
2	11(61.1)	-	7(38.9)
3	17(94.4)	-	1(5.6)
4	13(72.2)	-	5(27.8)
5	11(61.1)	-	7(38.9)
6	11(61.1)	-	7(38.9)
7	11(61.1)	-	7(38.9)
8	14(77.8)	1(5.6)	3(16.6)
9	8(44.4)	1(5.6)	9(50.0)
10	16(88.9)	-	2(11.1)

11	16(88.9)	-	2(11.1)
12	16(88.9)	-	2(11.1)
13	18(100.0)	-	-
14	13(72.2)	-	5(27.8)
15	9(50.0)	2(11.1)	7(38.9)
16	16(88.9)	1(5.6)	1(5.6)
17	15(83.3)	1(5.6)	2(11.1)
18	15(83.3)	1(5.6)	2(11.1)
19	16(88.8)	-	2(11.1)
20	13(72.2)	1(5.6)	4(22.2)
21	16(88.9)	-	2(11.1)
22	11(61.1)	2(11.1)	5(27.8)
23	14(77.8)	-	4(22.2)
24	15(83.3)	1(5.6)	2(11.1)
25	18(100.0	-	-
26	12(66.7)	-	6(33.3)
27	12(66.7)	-	3(16.7)
28	15(83.3)	-	2(11.1)
29	16(88.9)	-	2(11.1)
30	13(72.2)	4(22.2)	1(5.6)
31	15(83.3)	1(5.6)	2(11.1)
32	11(61.1)	2(11.1)	5(27.8)
33	10(55.6)	1(5.6)	7(38.9)
34	11(61.1)	2(11.1)	5(27.8)
35	18(100.0)	-	-

36	17(94.4)	-	1(5.6)
37	15(83.3)	3(16.7)	-
38	18(100.0)	-	-
39	10(55.6)	6(33.3)	2(11.1)
40	12(66.7)	4(22.2)	2(11.1)
41	18(100.0)	-	-
42	18(100.0)	-	-
43	16(88.9)	2(11.1)	-
44	15(83.3)	-	3(16.7)
45	14(77.8)	1(5.6)	3(16.7)
46	10(55.6)	1(5.6)	7(38.9)
47	12(66.7)	-	6(33.3)
48	13(72.2)	-	5(27.8)
49	15(83.3)	3(16.7)	-
50	16(88.9)	-	2(11.1)
51	15(83.3)	3(16.7)	-
52	18(100.0)	-	-

(Modified- M this implied only a change in expression to a suitable Nigerian expression e.g

‘spot’ inside of ‘find/see’) or Change (C-implied acomplete removal of a cultural

inappropriate term e.g a golf stick, to simply a stick)

Analysis of the 23 parents reported IQI responses as a questions inventory, showed a mean score of 1.72 from the 52 questions. Table 5.5.

Table 5.5: Subsections of the parent administered IQI and mean indicating the presence or absence of CVI/PVD

Domain	N	mean	SD
Visual Field	23	1.67	0.89
Perception of Movement	23	1.40	0.94
Visual guidance of Movement	23	1.76	0.96
Visual Search	23	1.89	1.22
Visual Attention	23	1.75	0.82
Recognition and Navigation	23	1.40	0.84
Mean Total	23	1.72	0.78

The corresponding IQI visual support strategies were chosen by parents upon identification of IQI questions that was responded to as sometimesor often. Parents identified the most important 3 visual support strategies to them. These strategies were explained to parents in details using relevant cultural and linguistic terms.

1.5.6 Observations

The result of nine hours of observations of three children randomly selected from the 23 children who presented with CP at their homes showeda discrepancy between parent IQI and observer's reports and scores with regards to IQI questions that the observers could make without asking parents for answers. Therefore, responses for all the questions on the IQI forms could not be obtained.

Child 1: Nine IQI observations were made by the KI; four questions were answered differently by

the carers from what the observer noted. These were in the areas of perception of movement (2), visual search (1) recognition and navigation (1). Parent reports of nine items showed a reported mean of 0.33 and an observed mean of 3.4

Child 2: Nine IQI observations were made by RD. All nine carers' reports appeared to differ from observation. The discrepancies were noticed in thedomains of perception of movement (3), visual search (3), visual attention (2) and recognition and navigation (1). Parental reports of nine questions showed a reported mean of 1.6 compared to the observed mean of 3.3.

Child 3: Six observations were made by the KI, and all six differed from parental reporting. These were in the areas of perception of movement(1), visual guidance of movement visual attention, (1) recognition and navigation (3). Parent report of six questions showed a reported mean of

1.0 compared to an observed mean of 3.3. This shows consistent underreporting and under recognition of visual perceptual problems by parents.

Presented below is one narrative report of observations:

"Observer RD asked for some water to drink. The mother called the index child and asked him to go to the kitchen to get RD water to drink.The child brought a glass and was asked to pour the water into it. He poured the water into the glass and filled it to overflow. The mother screamed angrily 'are you not looking'! The child turned to her, laughed and looked at her angry face and didn't appear to recognize the annoyance". This suggests possible abnormality in facial expression recognition and perception of movement.

The combined results from the IQI qualitative and quantitative studies were used to generate the IQI version 5, a revised package of 52 questionswith no questions expunged however, 22 questions were left in their original form, cultural and linguistic modification were done to 30 questionsand changed to suit those in Calabar municipality area of Cross River State in Nigeria and to be used

for a larger population survey. Appendix 4.

1.5.7 Result of PedQL

A total of 36 children were identified in the community. Thirty-four (34) children were brought. There were 21 (61.7%) males and 13(38.3%) females, between the ages of 4 to 15 years old. The mean age of the children brought was 8.6. years. There were 23 children with CP but 22 (64.7%) children with CP participated in the ocular examination. The mean age of the children with CP was 7.0. years. There were 10 (45.5%)males and 12(54.5%) females. The GMFCS levels 1-5 is described as follows: I- 4(18.8%); II-7(31.8%), III-2(9%), IV-8(36.8%), V-1(4.5%). Children where followed up after 2 weeks of administration of the intervention. There was no significant difference between the baseline and 2 weeks after scores in quality of life with the generic outcome. However, the PedsQL 3.0 CP showed a significant improvement in quality of lifein the subscale of school functions, while the subscale of pain showed a reduced quality of life. Table 5.6

Table 5.6: Description of the total mean scores on the PedsQL Generic

PedsQL generic Dimension	Baseline mean	Follow up mean	Difference	Improved /Worse	PValue
Physical	56.6	60.0	-0.528	Worse	.603
Emotional	64.3	62.5	.302	Worse	.766
Social	59.5	65.4	-1.357	Improved	.190
School	39.2	45.2	-0.607	Improved	.544

However, the CP results show a reduced quality of life from pain and tiredness. Table 5.7.

Table 5.7: Description of the difference in mean scores on the PedsQL CP module

PedQL CP Dimension	Baseline mean	Follow up mean	Difference	Improved/Worse QOL	P value
Daily activities	51.6	52.2	-0.12	Improved	0.9
School activities	27.6	49.6	-2.87	Improved	0.011
Movement and balance	56.1	65.0	-1.5	Improved	0.14
Pain & hurt	68.0	59.2	2.2	Worse	0.04
Fatigue	68.4	61.8	1.6	Worse	0.12
Eating	66.3	73.9	-1.1	Improved	0.28
Speech& Communication	51.1	47.0	.418	Worse	0.68

5.5 Discussion

The original version of the IQI can be culturally and linguistically modified to suit a particular population. The visual support strategies/interventions are explained in detail to the families and therefore the visual support strategies were not priority within the short time available for the study to conduct a translation/adaptation process.

For the successful performance of the Insight Question Inventory (IQI), parents and carers need to recognize and report abnormal behaviors related to CVI and PVD in their children. Even though the concept of the use of the IQI for identification of children with CV/PVD, was accepted and understood by most parents, the study shows that there may be the tendency to under report CVI/PVD using this instrument, because of under recognition of related behaviors by carers compared to the professional observers. Under-recognition and under-

reporting of visual problems may affect identification of CVI/PVD in children concerned. Furthermore, the tailored visual support strategies maybe few sincethese are only given in response to questions which are answered positively. It may tend to reduce the number of VSS given to each family, in direct response to the questions inventory response. However, the identification of the few that can be identified will at least be tailored to the behaviors that the parents are most aware of and concerned about, and small numbers of strategies may be easier to memorize and implement. Soon this basis despite evidence of under-reporting in IQI by carers we decided that it was worthwhile to proceed to the full trial.

Some questions and corresponding strategies 3, 5,15, 22,33,34 might have been expunged from the final version based on the results of careers/parent's comprehension. These questions had a poor response and referred to the use of computers. However, because of the diverse nature of participants and the level of education and exposure, the committee decided to retain all questions for the randomized trial. As new and locally suitable descriptions of CVI/PVD in children from Nigeria are identified over time, the set of questions in the modified Nigerian IQI inventory and visual support strategies might be expanded, while less salient ones removed.

A weakness of our study was that we did not take account of cultural variation in response to the Likert scale.(105)

The PedsQL CP module was shown to be feasible in assessing the quality of life in this children with CP. The pilot study did not show overall significant changes in PedsQL scores but there was an improvement in school related activities and a reduction in areas of pain and tiredness got worse; we therefore learned to explain to careers not to overtire children in practicing these strategies. We decided that the post intervention assessment could be done 6

weeks after the intervention to enable parents remember more strategies, as 3 months and 6 months maybe too long. Also the strategies should be administered in a prescriptive manner at least 3 times daily, to encourage parental motivation, focus and to remind parents to perform the strategies, but advising on the danger of causing pain and tiredness.

Acknowledgments

We acknowledge the key informants who participated in this study. Competing interests

The authors declare that they have no competing interests.

6 Clinical features and aetiology of cerebral palsy in children from Cross River State, Nigeria

Authors

Roseline Duke 1,2, Chimaeze Torty 3, Soter Ameh 4, Min J Kim1, Nnena Eneli 5, Ani Onyedikachi 5, Ada Aghaji1, Katie Burton 6, Richard Bowman 1

Affiliation

1London School of Hygiene and Tropical Medicine, International Centre for Eye Health, UK

2 University of Calabar Teaching Hospital, Calabar Children's Eye Centre, Department of Ophthalmology, Calabar, Cross River State, Nigeria.

3 University of Calabar Teaching Hospital, Department of Paediatrics, Neurology Unit, Calabar

4 University of Calabar Teaching Hospital, Department of Community Medicine, 5 University of Calabar Teaching Hospital, Department of Physiotherapy, Calabar 6 Cambridgeshire Community Services, UK

* Corresponding author: Roseline E Duke roseline.duke@unical.edu.ng

Calabar Childrens Eye Centre, Department of Ophthalmology University of Calabar Teaching Hospital, Calabar Cross River State, Nigeria

Key words: cerebral palsy, profile, children, aetiology

1.6 Chapter Abstract

Objective

There are few studies on cerebral palsy (CP) in African children and our study aimed to describe the aetiology, characteristics and severity of CPin children from Nigeria.

Design

A population-based study using key informant methodology (KIM) was conducted as part of a clinical research trial. Children aged 4-15 yearswere clinically assessed for CP.

Results

The estimated prevalence of CP using KIM was 2.2/1000 children (95% CI 2.7- 4.7). 388 children were diagnosed with CP, with Gross Motor Function Classification System Grade 1 in 70(18.1%), II in 156(40.2%), III in 54(13.9%), IV in 54(13.9%), V in 54(13.9%). CP types were spastic 70% (n=271), with 60% bilateral and 40% unilateral, ataxic 38(9.8%), dystonic 18(4.6%) and choreoathetoid 29(7.5%). 300/388 (77.3%)had manual ability classification scale (MACS) of level 1-3 and of 88(22.7%) of level 4-5. Malnutrition was seen in 200(51.5%). Neonatal morbidity, caused by birth asphyxia, hyperbilirubinaemia and clinical congenital rubella syndrome were identified as preventable causes of CP. Conclusion

CP in this population is similar to that found in other low-and-middle income (LMIC) countries. Some aetiologies identified were preventable.Prevention and management strategies for cerebral palsy designed for LMIC are needed.

1.7 Introduction

Cerebral palsy (CP) is a leading cause of childhood disability across the world with significant impact on function and development. (138) CP refers to a heterogeneous group of conditions involving permanent non-progressive central motor dysfunction that affects muscle tone, posture,and movement.(139, 140) The prevalence of CP is approximately 2- 3.5/1000 live births in high-income countries (HIC),(141) with a higher estimated prevalence of 2-10 per 1000 children in Africa.(29, 40) Population based studies on prevalence of CP in Africa are scarce.(29)

Several factors may influence the prevalence of CP in Africa, including the level of maternal or child healthcare and the neonatal morbidity ratesin study areas.(31, 34) CP results from a variety of insults to the immature, developing brain. The aetiology is multifactorial, may depend on socioeconomic factors. Prenatal and perinatal risk factors in HIC include low birth weight, preterm birth, and multiple pregnancy. (142) In LMICthey include maternal infections, neonatal jaundice, neonatal convulsions and infections, birth asphyxia and increasingly, prematurity.(31, 40) There is an increasing recognition of the importance of determining the impact of neonatal morbidity on long-term impairment in LMIC.(143) The aim of this study was to describe the prevalence, characteristic features of CP including the aetiology and severity in this population of Nigerian children, to guide service planning and prevention strategies.

1.8 Methods

1.8.1 Setting

A cross sectional study was conducted in Cross River State (CRS) in Nigeria between December 2017 and July 2018 in three districts (Southern,Central and Northern). These are made up of 18 local government areas (LGA), that cover an area of 20,156Km. The 2016 population projection for CRS was about 3.87million with about 1.1million children aged less than 14 years. Nine communities in three local government areas including; Odukpani (3 communities out of 13, Bakkasi (4 communities out of 13) and Ogoja (2 communities out of 10) with an estimated population of 150,000 children were not accessible for security reasons. The demographic and health data for Nigeria report that children between the ages of 5-14 years' account for 29.2% of the total population.(144) The tertiary hospital in CRS is located in the southern senatorial district. Two local government areas in the southern senatorial districts are urban and others rural.

1.8.2 Patients

Children with possible cerebral palsy from the 9 accessible regions of CRS were identified initially through key informant methodology. The key informant methodology (KIM) was chosen recognising its effectiveness for identifying physical impairment compared to household survey

in LMIC.(36) Key informants (KI) were local volunteers, all of them secondary school graduates, non-medical personnel, who lived and/or worked in their local community for at least 1 year, who knew the local context and community. They were identified in each community by the

village council and were trained to identify children with CP by using a locally produced picture manual developed specifically for the study for case identification with description of CP, and supplemented by the Ten Question Questionnaire to identify suspected cases.(145) These KI weregiven day-long structured training on CP and group sessions on awareness raising and disability-specific information using flip chart illustrationsand role play with guidance led by the principal investigator. The children identified by the KI were offered a consultation by a paediatric neurologist to provide an opportunity for diagnosis, treatment and referral of their child. Various search methods were used including house to house visits, school and church visits, also village and market place announcements. Cases suspected to have CP by the KI were brought to a primary health centre in each LGA.

Team members were trained in using research protocol form (i.e., data collection form which included sociodemographic details, mothers' antenatal history, prenatal perinatal and post neonatal factors.). All the children referred by the KI were assessed to determine if they met theinclusion criteria for the CP register. Families were provided with appropriate advice, information and counselling, referral services and intervention where appropriate.

Children aged between 4 and 15 years of age, who were confirmed to have CP from history and clinical assessment by a paediatric neurologistwere included in the study. Informed consent was taken. Children who had other motor disorders apart from CP and children outside the age criteria were excluded.

This study was part of a clinical trial and considering the population size for children as 1.1M, a 10% non-response rate, and an assumedprevalence of CP of 2.9 per 1000 (0.0029), a sample size of 370 would yield a prevalence precision level of 0.58%.

1.8.3 Cerebral palsy assessment

The diagnosis of CP was based on the history, and clinical examination through a neurological examination in line with international criteria.(139) CP was characterised clinically and by descriptions of the predominant motor pattern. Functional status was categorised with respect to motor activity using the gross motor function classification system (GMFCS) for CP and the manual ability classification system(MACS).(45)

1.8.4 Aetiology

The etiology for each case was determined clinically through caregiver interview including obstetric history, the child's prenatal, perinatal and post neonatal history, and determined by the physician's clinical examination where possible. Prenatal etiology might be indicated by an abnormal antenatal history including maternal drug or alcohol use, inconsistent or absent antenatal care, gestational diabetes, maternal infections,multiple gestation and febrile maternal illness. The perinatal period was defined as from 22nd week of gestation until 4 weeks after birth to allow

for uncertainty about timing of events after birth and include events related to birth.(146) An abnormal perinatal period was defined as any of the following: the need for a non-elective Caesarean section, preterm delivery (defined as babies born alive before 37 weeks of pregnancy), assisted delivery (vacuum or forceps), birth asphyxia (clear recollection of child not breathing or gasping at 5 min or a history of resuscitation), instrument assisted delivery and antepartum haemorrhage; and, post-natally, a child in whom there was an incident of any of the following: anyof neonatal convulsions, neonatal jaundice, infection and admissions for whatever cause to the neonatal unit. The aetiology of neonatal jaundicewas not investigated however, neonatal jaundice was felt to be significant and then referred to as hyperbilirubinemia in this study if poor suck was noted and the child was hospitalized with jaundice. Post-neonatal causes

were from the period following 4 weeks after birth and included malaria with seizures or coma, meningoencephalitis and identified HIV infection. The standard definitions for preterm birth, prolonged labour and microcephaly were used.(147) Clinical congenital rubella syndrome was diagnosed by signs of any two classic features without a more plausible aetiology (congenital heart disease, microcephaly, cataract or glaucoma, deafness and pigmentary retinopathy).(148, 149)

6.3.4 Statistical analysis

Statistical analyses were performed using Stata 15 (Stata Corp LP, College Station, TX). Descriptive statistics were reported using means and SD for normally distributed variables, medians and interquartile ranges for non-normally distributed continuous variables, and frequencies andpercentages for categorical variables. Comparisons between categorical variables were performed using a chi-square test or Fisher's exact test,and t-test was used for continuous variables. The estimated prevalence of children with CP in this population was derived by dividing the number of cases confirmed to have CP 4-15 years by the estimated total number of children aged 4-15 years in the population from the communities included in this study. Significance level was set at $P < 0.05$. Variables of interest occurred in at least 30% of subjects and missingdata were excluded.

Ethical approval

The study was approved by the London School of Hygiene and Topical Medicine and the Cross River State, health research and ethicscommittee with number CRS/MH/HREC/015/Vol.211.

1.9 Results

Key informants identified 1024 children, 293(28.6%) were not brought for examination, 344(33.6%) did not have CP and 388(37.9%) were confirmed to have CP. The estimated population prevalence of CP in children between 4-15 years using the KIM is 388/171200(2.26 CI 2.04-2.50) per 1000 children.

Socio-demography

There were no missing data in the variables of interest.

Table 6.1 describes the socio-demographic information of the children with CP (n=388). The mean age was 9.2 years (Standard deviation (SD) 4.0). One third of children were first born 133/388 (34.3%). Less severe GMFCS was seen in the older age group compared to the younger group($p<0.001$).

Table 6.1: Sociodemographic characteristics and secondary conditions of children with cerebral palsy (n=388)						
Characteristic		N	%	GMFCS 1,2,3, (n, %)	GMFCS 4,5 (n, %)	P value
Total		388	100	280(72.2)	108(27.8)	
Residence						
	Urban	44	11.3	33 (11.8)	11 (10.2)	0.656
	Rural	344	88.7	247 (88.2)	97 (89.8)	
Demographic						
Sex						
	Male	229	59.0	168 (60)	61(56.5)	0.528
	Female	159	41.0	112 (40)	47 (43.5)	
Age						
	Mean (SD)	9.2	4.0			
	Median (IQR)	9	6,13			
	<9 years	176	45.4	108 (38.6)	68 (63.0)	<0.001
	9+ years	212	54.6	172 (61.4)	40 (37.0)	

Standard vaccines (Bacillus Calmette-Guerin, Oral Polio Vaccine, Diphtheria Pertussis Tetanus, Haemophilus Influenza, Measles) were not administered at all in 3/388(1%) children. In 63/388(16.24%) children, uptake of immunization was incomplete, and this rate was significantlydifferent between the rural and urban areas. Six (2%) children had not received doses 1 and 2 of Vitamin A supplementation. Rubella immunization is not given in Nigeria.

CP description

Spastic CP was the most common type (271/388,69.9%), and was bilateral in 163/271(60.2%) and unilateral in 108(39.8%). Table 7.2 describesthe type of CP and anatomic distribution.

Table 6.2: Distribution of cerebral palsy subtypes in study participants

Subtypes	N(%)
Unclassifiable	32(8.25)
Spastic	271(69.85)
Bilateral Spastic	163(60.2)
Unilateral spastic	108(39.8)
Dystonic	18(4.64)
Choreoathetoid	29(7.47)
Ataxic	38(9.79)

Levels of GMFCS and type of CP are shown in Table 6.3.

Table 6.3: Distribution of GMFCS levels and CP subcategories in study participants

GMFCS level	N (%)	
		Ambulatory
I	70 (18)	280 (72.2)
II	156 (40)	
III	54 (14)	
		Non ambulatory
IV	54 (14)	108 (27.8)
V	54 (14)	

Neurologic subtypes	Total	I
Total		70
Unclassifiable	32	17 (53.1)
Spastic	271	26 (9.6)
Dystonic	18	12 (66.7)
Choreothatetoid	29	3 (10.3)
Ataxic	38	12 (31.6)

GMFCS levels were categorized as ambulatory (Levels I–III) (n=280, 72.2%) and non-ambulatory (Levels IV–V) (n=108, 27.8%). MACS levelswere: I 178 (45.9%); II 75(19.3%); III 47(12.1%); IV 80(20.6%); V 8(2.1%). Children with MACs levels 4-5 were more likely to be non- ambulatory ($p<0.001$).

More children with spastic CP and fewer with choreoathetoid CP had higher levels of motor difficulties as on the GMFCS (Table 6.4).

Table 6.4: Pearsons X^2 test, neurologic subtypes of cerebral palsy(CP) vs GMFCS level

	N (%)	GMFCS 1,2,3 (n, %)	GMFCS 4,5 (n, %)	X^2	P value
Total observations	388 (100)	280 (72.2)	108 (27.8)		
Type of CP					
Spastic CP	271 (69.9)	178 (65.7)	93 (34.3)	18.8	<0.001
Ataxic CP	38 (9.8)	36 (94.7)	2 (5.3)	10.7	<0.001
Dystonic CP	18 (4.6)	16 (88.9)	2 (11.1)	2.6	0.105
Unclassifiable CP	32 (8.3)	27 (84.2)	5 (16.6)	2.6	0.08

Choreoathetoid CP	29 (7.5)	23 (79.3)	6 (20.7)	0.8	0.372

Aetiology

Timing of causation was deduced by the physician to be prenatal in 8/378(2.1%), perinatal in 233/378 (61.6%), and post neonatal in 137/378 (36.2%) and is detailed in table 6.5. Fifty-nine out of 315(18.7%) developed hyperbilirubinemia and 29 of these children had choreoathetoid CP; 10/29(34.5%) out of the total number with choreoathetoid CP were first born children, which is not significantly different from the overall groupchildren with CP. Symptoms of birth asphyxia, occurred in 118/388 (30.4%). Eight children (2%) met the criteria for clinical congenital rubellasyndrome (CRS). Premature birth was reported in 20/388(5%) children.

Table 6.5: Distribution of aetiology by neurological subtype from clinical assessment

Aetiology	Total	Spastic	Choreoathetoid	Dystonic	Unclassified	Ataxic
		N (%)	N (%)	N (%)	N (%)	N (%)
Perinatal	233	163 (61.3)	28 (96.6)	8 (47.1)	20 (64.5)	14(40.0)
Prenatal	8	6 (2.3)	0 (0)	0 (0)	1 (3.2)	1 (2.9)
Post-neonatal	137	97 (36.4)	1 (3.4)	9 (52.9)	10 (32.3)	20 (57.1)

1.10 Discussion

The prevalence estimate of CP in this population was close to other estimates from similar populations in Africa.(29) The KIM methodology waschosen because it offers a low-cost, relatively rapid method to identify children with CP in a LMIC, as extensively reported in Bangladesh.(36) The clinical diagnoses of non-CP cases were also similar to other African studies.(Supplemental Table)(29) Our estimated prevalence was slightly lower than the Ugandan study (2.2 vs. 2.7 per 1000) and the recent study from Bangladesh (2.3 vs 3.4/1000),(33) but our study involved older children and early mortality may therefore act to reduce our recorded prevalence. We are also aware of stigma associated with public appearance of children with CP, which could result in under reporting, through KIM methodology versus a door to door sampling method.(51) Apotential advantage of KIM methodology is that it also provides the opportunity to plan interventions using a community-centred approach which encourages access to health services

CP prevalence in this setting, may be causally related to other indicators of poor health in the country such as the high incidence of maternal andneonatal morbidity.(143) In this population, the aetiology of CP appears to be mixed, with a proportion from preventable causes. In LMIC, hyperbilirubinemia with bilirubin encephalopathy, is a significant cause of neonatal neurodevelopmental morbidity and is a priority for effectiveintervention strategies.(38) A number of studies have reported that many mothers delay seeking timely and appropriate care.(150) Risk may be

higher in a first time pregnancy. Strategic health education for mothers on early identification of symptoms of hyperbilirubinaemia, may promoteearlier presentation and effective intervention. Symptoms of birth asphyxia were seen in about one third of cases of CP which may be

significant suggesting improved antenatal and perinatal care could improve outcomes. However, the direct contribution of adverse intrapartum and obstetric events to overall CP rate is thought to be less than suggested by overall occurrence of birth asphyxia symptoms The causal pathway model highlights that there are likely to be a networkof earlier risk factors for cerebral palsy such as maternal infection and poor placental function which may lead to later presentation with symptoms of birth asphyxia and CP. Understanding the network of factors that contribute to the causal pathway may identify points for intervention and promote prevention rather than ameliorating the injury. (151, 152) Prenatal aetiologies are difficult to ascertain accurately from observational studies, and true ascertainment about whether the primary cause is pre or perinatal is challenging in this type of study.(140) However recognising contributory causes in this setting is important for targeting prevention strategies. Another likely preventable cause was congenital rubella syndrome which is one of the leading vaccine-preventable cause of birth defects. Vaccines have been shown to have significantly decreased the incidence of congenital rubella syndrome in HIC.(153) We noted that a small percentage of children were either not immunized at all (1%) or did not have full immunization uptake (16%), similar to another studies from Australia.(116) Further research is needed into the factors that influence uptake of full immunization in children with CP and to identify barriers and enablers to accessing vaccination. As neonatal care in LMIC improves, multiple pregnancies and prematurity may become more common as associated causes of CP.

The physical characteristics of CP in this population showed similar distribution in type, GMFCS, MACS, with that seen in other LMIC.(116)Less severe GMFCS was seen in the older age group compared to the younger, suggesting a higher mortality at an early age in those with the highest GMFCS as seen in similar LMIC studies.(30, 33)

The study was conducted in one state in the country which has a higher socioeconomic index

compared to the rest of the country, and the prevalence of CP was an estimate by the KIM, making it difficult to generalize for the whole country or region. Likewise, security problems restricted access to 9 communities in the study area which could also introduce bias, and reduced access in these troubled areas to maternal and neonatal healthcare could also affect prevalence of CP. Sources of inaccuracy include KIM methodology (selection bias) and likely stigma limiting the number of children brought forward for assessment. Judgement about aetiology is likely to have been affected by recall bias. In spiteof these limitations, the study used trained personnel as interviewers, international protocols and classifications and represents a large sample of children with CP.

Conclusion

The profile of CP in this community-based study in Nigeria is one of mixed aetiology, including preventable neonatal conditions such as hyperbilirubinemia, congenital rubella, and likely birth asphyxia. There is a need to focus on primary prevention strategies for these commoncauses of neonatal morbidity and to provide holistic care.

Data statement

The protocol and data are available from the authors upon reasonable request and with the permission of the London School of Hygiene andTropical Medicine.

Funding

The Queen Elizabeth Diamond Jubilee Trust/Common Wealth Eye Health Consortium funded the research grant number [LSHTM ITCRZ6814].

Acknowledgments

We acknowledge all the carers of children with CP and the key informants who participated in this

study.

Competing interests

The authors declare that they have no competing interests.

Pattern of comorbidities in school-aged children with cerebral palsy in Cross River State, Nigeria

Authors

Roseline E Duke FWACS 1,2, Chimaeze Torty FWACP 3, Uche Okorie OD, Min J Kim MPH1, Nnena Eneli BSc4, Ukam Edadi MSc6, Kathryn Burton MD5, Cally Tann PhD7 Richard Bowman MD1

Affiliation

1London School of Hygiene and Tropical Medicine, International Centre for Eye Health, UK

2 University of Calabar Teaching Hospital, Calabar Children's Eye Centre, Department of Ophthalmology, Calabar, Cross River State, Nigeria.

3 University of Calabar Teaching Hospital, Department of Paediatrics, Neurology Unit, Calabar

4 University of Calabar Teaching Hospital, Department of Physiotherapy, Calabar

5 Cambridgeshire Community Services, UK

6 Cross River State, Primary Health Care Development Agency, Calabar

7 Clinical Research Department, London School of Hygiene & Tropical Medicine, London, UK

Email address of authors

Roseline E Duke:dr.roseineduek@gmail.com

Chimaeze Torty: drchimatorty@gmail.com Uche Okorie: ucheman_b@yahoo.com

Min J Kim: Min.Kim@lshtm.ac.uk

Nnena Eneli: nenaenelii@gmail.com: Ukam Edadi: timukam@gmail.com

Kathryn Burton: kjburton@doctors.org.uk Cally Tann: cally.tann@lshtm.ac.uk

Richard Bowman: richardbowman493@gmail.com

* Corresponding author: Roseline E Duke

Calabar Childrens Eye Centre, Department of Ophthalmology University of Calabar Teaching Hospital, Calabar

Cross River State, Nigeria dr.roselineduke@gmail.com

Key words: cerebral palsy, comorbidity, school attendance, children

Abstract Objective

To describe the pattern of comorbidities in school-aged children with cerebral palsy (CP) and to identify which, if any, were associated with poor school attendance.

Methods

A cross-sectional study, using the key informant methodology, between December 2017 and July 2018 was conducted in Cross River State, Nigeria. Assessments, confirmation of CP and identification of systemic comorbidities using standard tools and questionnaires were performed. Children confirmed to have CP between the ages 4 to 15 years were included.

Results:

Three hundred and eighty-eight children were confirmed to have CP, 59% males. The mean age was 9.2 years ± SD 4.0; 28% were non- ambulatory (gross motor function classification system (GMFCS) level IV-V) and spastic CP was seen in 70%. Comorbidities included Speech impairment 85%, feeding difficulties 86%, and swallowing difficulties 77%, learning difficulties 88%, abnormal behaviour 62%, visual acuity impairment 54%, objective perceptual visual disorders 46%, communication difficulties 45%, epilepsy 35%, hearing impairment 12% and malnutrition 51%. Learning difficulties (OR 10.1, p<0.001; CI: 3.6-28.1), visual acuity impairment (OR 2.8, p=0.002; CI: 1.5-5.3), epilepsy (OR 2.3, p=0.009;

CI:1.2-4.3) manual ability classification scale 4-5 (OR 4.7,p=0.049; CI:1.0-22.2) and CP severity (GMFCS V-VI) OR 6.9 p=0.002, CI: 2.0-24.0.) were seen as increasing the likelihood of poor school attendance.

Conclusion

Comorbidities were common, and some were associated with limited school attendance. A multidisciplinary tailored approach to care, with application of available therapeutic interventions for comorbidities is suggested. This may be useful in reducing barriers to school attendance.

7.1 Introduction

Cerebral palsy (CP) is a leading cause of childhood disability across the world with significant impact on function and development.(154) The definition of CP highlights frequent association with disorders of sensation, perception, cognition, communication, behaviour, with epilepsy and secondary musculoskeletal problems.(139) Co-morbidities affect the overall health and quality of life in children by determining their participation in most aspects of life including schooling.(98) Secondary conditions (e.g. joint contractures and malnutrition) occur and are preventable. Children with more severe levels of gross motor dysfunction present with comorbidity more frequently.(30, 114) It is estimated that the majority of children with disabilities in Africa do not go to school at all,(155) and of the 72 million primary aged children worldwide that are out of school, one third have disabilities.(156) Few reports describe school attendance and educational attainment in children with CP inLow and Middle Income Countries (LMIC).

Children with CP and disability in general face barriers and challenges to education.(155) Barriers to attending school are commonly describedthrough the 'social model' of disability, that is about the way the society responds to children with disability. The common ones are physical, social and financial. (157) This 'social model' of disability differs from the 'medical model' which sees people with disabilities as having a problem that needs to be managed, changed and/or adapted to circumstances.(158)

Co-morbidities may contribute to these barriers. Little evidence exists as to whether and which comorbidities are significant barriers to participation in schooling. Identification of these comorbidities may be important to attain effective individualized support measures for education.

In Nigeria, there are strong regional disparities in education and socioeconomic indices, with the

southern region of the country (where Cross River State is situated) performing better in indices, including of health, education and the millennium development goals.(159) In the general population of children in Nigeria, the gross enrolment rate in elementary school is 68.3% , gross enrolment rate in lower secondary is 54.4% andupper secondary 68.9%(160) With regards to malnutrition , 37 per cent of children are stunted and in addition 18 per cent of children suffer fromwasting while twenty-nine per cent of children are underweight.(161)

The aim of this study was to describe the comorbidities seen in children with CP in this community –based Nigerian population, and to identifywhich comorbidities were associated with poor school attendance.

7.2 Methods

7.2.1 Study setting/context:

A population based cross sectional study was conducted in Cross River State (CRS) in Nigeria between December 2017 and July 2018 and hasbeen reported in other articles.(95, 162, 163) Primary and secondary education enrolment is compulsory; however, it is paid for by the government for children in the primary school, while the government subsidizes the examination fees for the secondary education. Located in every village is a primary school and in every local government area there is a state government secondary school and a mission secondary school. Enrolment into kindergarten is from 3 years old.Secondary school education is divided into junior secondary and senior secondary schools.(164) There are three government special education schools in the state for deaf, mute and visually impaired children.

7.2.2 Sample size

Existing data from this world region suggest a population prevalence of CP of 2.9 per1000

children and about the same estimate for associated comorbidities.(29, 165, 166) Considering the population size for the total number of children aged 4-15years in Cross River State as 1.1M,(144)a 10% non-response rate, a sample size of 370 would be sufficient to estimate a prevalence of 2.9/1000 (i.e. p=0.29%) and associated comorbidities(29) with a level of precision of ±0.58%.

7.2.3 Sampling strategy

In the absence of a cerebral palsy registry, the key informant methodology (KIM) was selected as the most cost effective sampling strategy to identify children with CP and other disabilities in our circumstances. Several researches have validated this method (33, 167-169). The key informant methodology (KIM) was chosen, recognising its effectiveness for identifying physical impairment compared to household survey inLMIC. (36) The methodology used is referred to in other articles.(95, 162, 163)

Identified and referred children were then assessed first of all by a paediatric neurologist in the primary health centre to determine if they met theinclusion criteria for CP. Families were provided with appropriate advice, information and counselling, referral services and intervention where appropriate. CP was defined according to history and neurological examination in line with international criteria.(28) Gross Motor Function Classification Scale (GMFCS) was used to describe the severity of CP of gross motor impairment. These levels were categorized into ambulatory (Levels I–III) and non-ambulatory (Levels IV–V).(170) Validated existing questionnaires and tools were used such as the strength difficulties questionnaire, the manual ability classifications system and the communication function classification system.

The inclusion criteria included, children aged between 4 and 15 years of age at their last birthday, who were confirmed to have CP from history and clinical assessment by a paediatric

neurologist.(171) Exclusion criteria included children who had other motor disorders apart from CP andchildren outside the age criteria and those that refused to participate in the study.

7.2.3 Comorbidity case ascertainment

Comorbid conditions were confirmed by history, clinical and standardized evaluations. Comorbidities investigated included: epilepsy, hearingimpairment, feeding difficulties, swallowing difficulties, visual acuity impairment, objective perceptual visual disorders, abnormal behaviour, learning difficulty, speech impairment, communication difficulties and malnutrition. The Lea symbols cut-off point for screening preschool children of 0.8 was used as score for normal visual acuity and >0.8 were considered tohave visual acuity impairment. (172) Objective perceptual visual impairment was ascertained by a battery of tests.(95)

Hearing impairment was assessed using three-level voice test (for children able to participate) and was present when a child failed to respond tomid-level spoken voice in either ear. Learning difficulty was assessed, by clinical history, assessment and behavioural observation.(173, 174) The Communication Function Classification Scale (CFCS) assessed the full activity of communication in five levels between a familiar person and the child.(48) We referred to children as having communication impairment if CFCS was level 4-5. Speech impairment were defined as inability to create or form speech sounds.(175) Epilepsy was diagnosed on a history of having two unprovoked seizures >24 hours apart at any time from one month of age to assessment.(176) Feeding difficulties was based on the reported ability of the child to chew food and the need forfood to be cut up or mashed. Swallowing difficulties was defined as choking and coughing on food or drink based on parental report.(177) The manual ability classification system (MACS),(178) categorized as 1-3 and 4-5 as severe manual ability impairment. The strength and difficultiesquestionnaire(SDQ),(179) which is an emotional and behavioural screening questionnaire, was used through parent interview to describe any behavioural abnormality, emotional and conduct problems, hyperactivity, peer problems and

prosocial behaviours.(180) Behaviour disorder wasdefined using the total difficulties score from the Strength and Difficulties Questionnaire. Malnutrition was classified using the Centre for Disease Control (CDC) growth chart(for ages 2-19 years),(181, 182) and comprised of underweight or wasting, stunting and overweight. Severeacute malnutrition was defined as weight for height at least 3 SD below the reference median or mid- upper arm circumference less than 11.5cmfor children less than 5 years.(183)

Participation in mainstream schooling was recorded alongside whether children were at expected levels within the school programme by parent'sreport. Dropout in school was defined as the percentage of students failing to complete a particular school year or school program.

7.2.4 Statistical analysis

Statistical analyses were performed using Stata 15 (Stata Corp LP, College Station, TX). Descriptive statistics were reported using means, standard deviations, medians, interquartile ranges, frequencies and percentages. Comparisons between categorical variables were performedusing a chi-square test and logistic regression.

Comorbidity score was calculated by summation of the frequencies of the following comorbidities: feeding, swallowing, hearing, speech, learning, visual acuity impairments and objective perceptual visual disorders. In addition to; malnutrition, communication difficulties, epilepsyand abnormal behaviours. This was followed by the calculation of the comorbidity mean.

The Kruskal Wallis test was used to determine the association of comorbidity scores with the type of CP. Significance level was set at $P < 0.05$.Missing data if less than 30% were included as normal. The influence of co-morbidities and CP severity on school attendance was assessed in abivariate analysis. Correlations between factors predicting school attendance were the sought.

Multiple logistic regression models, adjusted for age and sex, were developed to identify factors associated with poor school attendance. Variables included in the regression models included systemic comorbidities and severity of CP. These were chosen based on biological plausibility and findings from previous studies. In addition, a no selection procedure was used to include other factors: variables significant at $p<0.2$ level, or not significant at $p>0.2$ but with an odds ratio between 0.5 and 2.0 in bivariate logistic regression were also included in the multivariate model. If 2 predictors showed a strong correlation with each other (0.7-1), then only one was included in the multivariate modelling.Age and sex were included regardless.

7.3 Results

A total of 1024 children were identified by the key informants, 343(34%) children referred did not have CP while (388/731(53%) were confirmed to have CP at that point in time. The mean age of the children with CP was 9.2 years (SD) ±4.0). There were 229 (59%) males and159 (41%) females. Carers reported seeking treatment for CP first in the hospital in 56.7%.

Ambulatory children (GMFCS I-III) made up 280/388 (72%) while GMFCS IV-V were 108(28%) of the children with CP. Spastic CP was themost common type (271/388,70%), and was bilateral in 163/271(60%) and unilateral in 108(40%).

7.3.1 Comorbidities

Comorbidity distribution are shown in Table 7.1

Table 7.1: Distribution of comorbidities and other variables in an unadjusted bivariate analysis showing predictors of school attendance(n=388)

Type of comorbidity	N (%)	Crude OR	95% CI		P value
Feeding difficulties	334(86)	0.06	0.01	0.3	<0.0001
Learning difficulties	342 (88)	15.8	6.8	36.7	<0.0001
Speech impairment	331(85)	2.6	1.5	4.7	0.001
Swallowing difficulties	299(77)	0.3	0.2	0.6	<0.0001
Abnormal Behaviour (Total difficulties score)	231(62)	1.2	0.8	1.9	0.355
Visual acuity impairment	209(54)	6.3	3.9	10.1	<0.0001
Communication difficulties (CFCS 4-5)	173(45)	4.8	2.9	7.8	<0.0001
Objective perceptual visual disorders	177(46)	5.2	1.2	22.8	0.027
Epilepsy	130(35)	3.0	1.8	4.9	<0.0001
Hearing impairment	46(12)	1.3	0.6	2.5	0.468
Malnutrition	200(51)	1.8	1.2	2.8	0.005
GMFCS IV-V	108(28)	16.1	6.3	40.8	<0.0001
MACS 4-5	88(23)	20.2	6.2	65.4	<0.0001

{OR >1 means significantly associated with poor school attendance, OR <1 means significantly associated with better school attendance}

Subcategories of abnormal behaviours included; Difficulties getting along with other children 240(63%), reduced kind & helpful behaviour 252(67%), hyperactivity and inattention 162(43%) and abnormal conduct 233(62%). Neonatal seizures 108/388(28%) (OR 4.4, 95% CI 2.8-7.1;

p<0.001), were four times more likely in children with epilepsy. Irregular antiepileptic medications were used in 7/130(5.4%) children whileothers used none.

Malnutrition was seen in 200/388(51%) and was associated with MACS 4-5, 62/88(70%); (OR 2.8,95% CI: 1.7- 4.7; p<0.001), GMFCS IV-V, 83/108(77%) (OR 4.6; CI:2.8-7.7;7p<0.01). Conversely a negative association was seen with both feeding, 161/334(48%) (OR 0.3; CI:0.2-0.7; P<0.001) and swallowing difficulties 140/299(47%) (OR 0.4; CI:0.2-0.7; p<0.001).

The comorbidity score showed a mean of 6·4 (SD 1·9; median 6; IQR 5,8), with the Kruskal-Wallis test showing a significant difference in the distribution of the co-morbidity scores between the CP clinical types ((X^2 (4) = 10.921, p <0·0275); Dystonic CP showed the highest number ofco-morbidities; 7.4 (SD 1·8 Median 7.5, IQR 6,9). Children with more than 5 comorbidities accounted for 65% of children and at least 1 comorbidity was seen in every child. (Figure 1 and supplementary material 1).

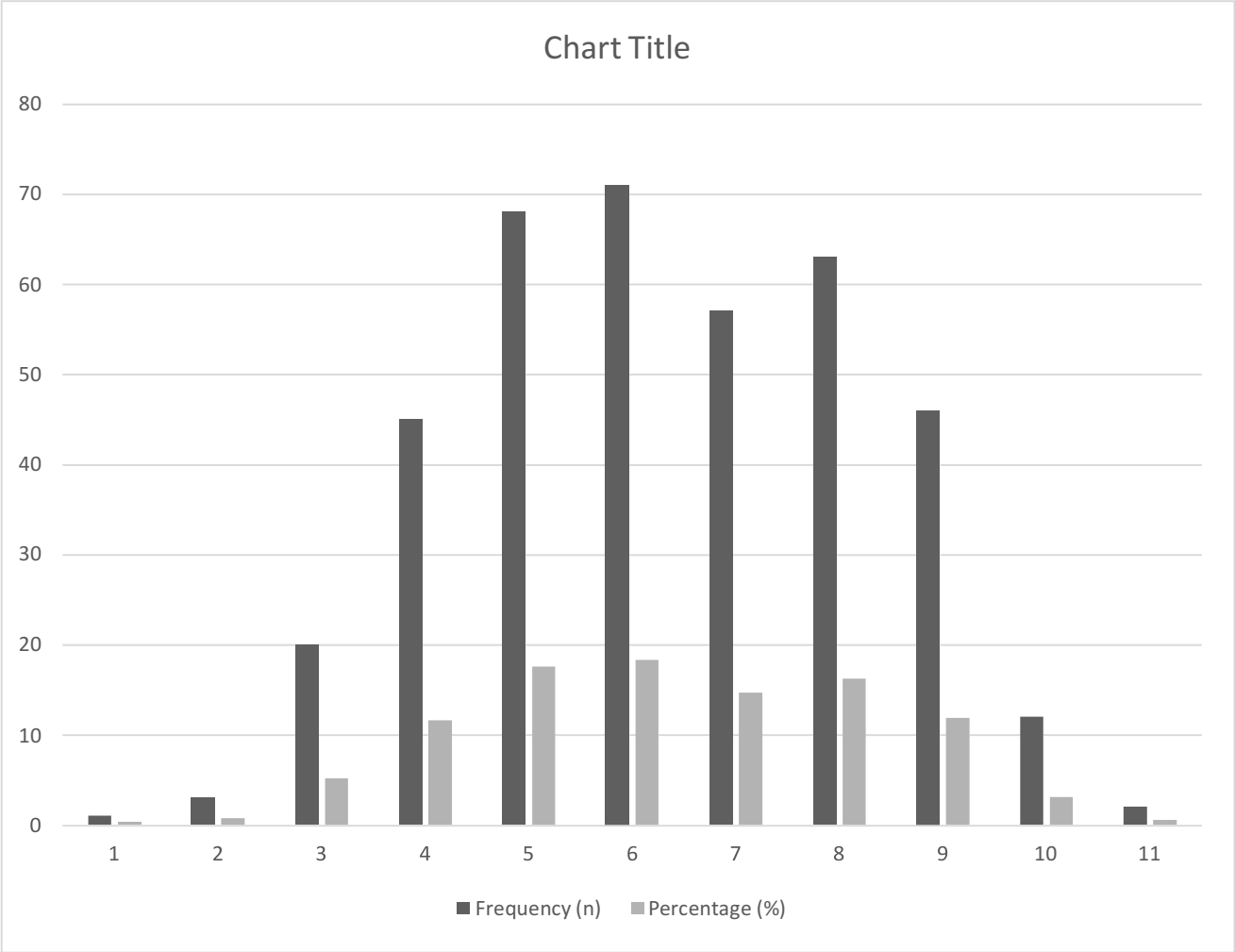


Figure 7.1: Number and frequency of comorbidities in children with CP (n=388)

Supplementary material 7.1: Number and frequency of comorbidities in children with CP (n=388)

No of comorbidities	Frequency (n)	Percentage (%)
1	1	0.26
2	3	0.77
3	20	5.15
4	45	11.6
5	68	17.53

6	71	18.3
7	57	14.69
8	63	16.24
9	46	11.86
10	12	3.09
11	2	0.52

Schooling

All the children recruited were of school age, 115/388(30%) had never attended school, and 145 of the 273(53%) who started school droppedout; of the 128/388 (33%) in mainstream school, for 124/128 (97%) children, parents reported that the children were behind in academic performance. As at the time of recruitment, two thirds of children were not in school (260/388, 67%).

Multivariate analysis

Of the reported bivariate predictors of school attendance only swallowing and feeding difficulties showed some correlation (R=0.7) henceswallowing difficulties was deleted from the multivariate model.

Age and sex adjusted multivariate analysis is shown in, table 7. 2.

Table 7.2: Multivariate logistic regression analysis of comorbidities and other factors predicting poor school attendance in children with CP(n=388)

Variables	Multivariate analysis			
Type of comorbidity	Adjusted OR	P value	95% CI	
Age > 9years	0.3	0.001	0.2	0.6
GMFCS IV-V	6.9	0.002	2.0	24.0
MACS 4-5	4.7	0.049	1.0	22.2
Feeding difficulties	0.1	0.039	0.02	0.9
Learning difficulties	10.1	<0.001	3.6	28.1
Visual acuity impairment	2.8	0.002	1.5	5.3
Epilepsy	2.3	0.009	1.2	4.3

7.4 Discussion

This population based study on children with CP from Nigeria a LMIC study, suggests evidence on specific comorbidities and their negativeimpact on school attendance which were independent of CP severity.

Previous studies on CP in children from Nigeria and LMIC have mostly been from facility based samples rather than the community. There haverecently been population-based studies from Bangladesh on prevalence and co-morbidity,(33) using the same methodology and from Uganda, on prevalence only.(29) Review of these, has highlighted there is a need for further population-based studies from other LMIC to understand cultural and geographic differences in the burden of co-morbidities that has differed across regions.(138). Furthermore, few studies in

LMIC are available on children with CP from community-based studies in relation to participation in schooling and most information regarding schooling,have been based on hospital samples.(30, 32)

Comparative studies have shown that the KIM can be used effectively to estimate a prevalence and identify associated comorbid conditions andpredictors. For instance, a large sample of children with CP with physical impairment have been identified through the key informant method inBangladesh.(33, 167, 171) However, the issues of stigma, difficulty with movement and poor expectations for treatment of the condition may have discouraged some parents from bringing their child for examinations.(51)

All the comorbid conditions in our study occurred in higher frequency than are reported in studies from High Income Countries.(184, 185) In theUgandan study, two comorbidities were reported (learning disability and epilepsy) in similarly high frequencies and this was from a hospital- based study where one might expect higher level of difficulties that had resulted in referral to hospital.(30) In comparison to Bangladesh, (33) the proportion with various co-morbidities was similar for some e.g. hearing impairment and higher in others e.g. visual acuity impairment.
Differences seen could well be related to different screening and assessment measures as well as reflecting population differences. Our studyalso included other areas of difficulty which can have a significant impact such as behaviour and malnutrition.

In our population, very few of the modifiable comorbidities, such as epilepsy, were receiving treatment. Feeding and swallowing difficulties were very common but were surprisingly associated with significant reduced likelihood of malnutrition compared with severe CP and manual ability 4-5, which both showed an increased likelihood for malnutrition. Similar studies,

(186, 187) have shown an association between feeding and swallowing problems and malnutrition,.(41) What may appear as a discrepancy may be as a result of the cultural feeding norms where from the age of above 4 years in these communities, feeding is communal with all the children in the household feeding from the same plate together,with the older child expected also to assist the younger children to obtain food from the plate. Children with severe CP and manual disability, aremost unlikely to compete with their normal peers for the food. Suggesting the aetiology of malnutrition may not only be as a result of the difficulties in swallowing or chewing in these communities in children above 4 years of age.

Similar to a population based study from Uganda,(30) children with dystonia had the highest mean comorbidity score. This may reflect moreglobal insult from underlying aetiology of CP e.g. neonatal encephalopathy in these children. Comorbidity is associated with worse health outcomes, more complex clinical management, and increased health care costs.(188) The relationship between the number and specific comorbidity per child and mortality in children with CP requires further investigation across LMIC.

The importance of participation in education by children with disability including CP has been reiterated by several organizations.(155). We found no child attending special education school in our study. A low prevalence was seen in another Nigerian hospital-based study where only 8% attended special education schools.(32) One of the major reasons identified by the earlier study for keeping the children away from school was fear of stigma and the assumption from family members that the children were not capable of learning.(32) It is possible that parents may have noticed some of the comorbidities but did not understand or assumed that they could not be addressed. The academic expectation of children with CP should be tailored and agreed with parents. Understanding the link between poor school performance and comorbidity would help towards more individualised child centred approaches of care. For example, the

consequence of untreated epilepsy could result in deleterious cognitive and behavioural consequences, (189) both of which could be ameliorated.

Significant determinants of poor school attendance in this population independent of the severity of CP based on ambulation and manual disability, are: epilepsy, learning difficulties and visual acuity impairment. Some of these have proven effective interventions when indicated,

(41) for example, the use of antiepileptic medication in some children and the use of spectacles in children with refractive errors and/or accommodative dysfunction which are known to be beneficial.(190, 191) Interventions may improve the quality of life, school participation, performance and favourable competition with their peers. (17) Apart from environmental and social interventions which addresses non- ambulation and manual ability, interventions to ameliorate comorbidities in children with CP towards improvement in schooling should be considered. Focus on the development of special education schools and complex facilities to improve capacity for clinical care, habilitation andeducation of children with more severe CP may be beneficial.

Children older than 9 years were seen to have a reduced likelihood of poor school attendance. There may be a link between mortality of children with CP and different age groups as well as between mortality and the development of adaptations in children with CP. These require further investigations in the implication for school attendance. A similar protective finding of significance was feeding difficulty; These was seen maybebecause of the ongoing school feeding programme where parents who are aware their children's malnourished state, but do not understand the cause of the malnutrition send their children to school to participate in the school feeding programme. More investigations would clarify reasons why this is seen.

Limitations of the study

Recruitment using key informant methodology is not as rigorous as door to door population surveys but is more practical and cost effective andhas been used for large and well analysed studies of CP in Bangladesh.(33, 167) Our KIM methodology has been based on the Bangladesh

model however, it may have missed children resulting in selection bias. For instance, it may have been that children with more severe and stigmatising CP were not brought thus possibly underestimating the degree of co-morbidity in this population and hence the public health significance. Identification of comorbidities in some areas relied on parental report with possible recall bias however, there were additional extensive professional assessments used to determine comorbidity. Lastly, our choice of cut-off values affects the frequency of co-morbiditiesand also most likely their impact as predictors of the outcome variables.

In spite of these limitations, the study probably reflects this country's best case scenario,(159) used a well-established sampling methodology,trained personnel as interviewers, international classifications, and it represents a large sample in a LMIC population.

Conclusion

CP severity contributes significantly to poor school attendance, hence the social model of care in disability should continuously be strengthened.However, the majority of school-aged children with CP in this large population-based study in Southern Nigeria showed a high prevalence of multiple, untreated co-morbidities highlighting CP in this population as a multimorbid condition which may be contributing adversely to school attendance. Some of these co-morbidities that present as barriers are modifiable and if properly managed, may have the potential to have positiveimpacts on school attendance.

United Nations Sustainable goal four is for quality education and over half of the children

who are not enrolled in school globally are in subSaharan Africa and Nigeria is by far the largest country in sub Saharan Africa. The sustainable goals emphasise the principle of not leaving anyone behind (universal coverage) and this study highlights that comorbidity is likely to be a major impediment to school attendance forchildren with CP in low and middle income countries which requires urgent attention.

Acknowledgments

We acknowledge all the carers of children with CP and

the key informants who participated in this study.

Conflict of interests

The authors declare that there is no conflict of interest regarding the publication of this paper.

Ethics approval and consent to participate

The study was performed in accordance with the Helsinki declaration and approved by the ethics committees of Cross River State and the London School of Hygiene & Tropical Medicine. A written information sheet was read out and informed consent was obtained from all subjectsunder 18, from a parent and/or legal guardian.

Children were referred for health care services as needed.

Funding: The Queen Elizabeth Diamond Jubilee Trust/Common Wealth Eye Health Consortium funded the research. Grant number [LSHTMITCRZ6814].

Availability of data and materials

The datasets used and/or analysed during the current study are available

from the corresponding author on reasonable requestAuthors' contribution

Conception and design of the work: Richard Bowman, Roseline E Duke, Kathryn Burton

Data collection: Roseline E Duke, Chima Torty, Uche Okorie, Ukam Edadi, Nnena Eneli; Data analysis and interpretation: Roseline E Duke, Min J Kim; Drafting the article: Roseline E Duke; Critical revision of the article: Kathryn Burton, Cally Tann , Richard Bowman; Final approvalof the version to be published and agreement to be accountable for all aspects of the work: Roseline E Duke, Chima Torty, Uche Okorie, Min J Kim, Kathryn Burton, Richard Bowman.

Acknowledgements

The key informants, Caroline Arthur and Totsman Daniel the field coordinators, are acknowledged.

List of abbreviations

CP Cerebral palsy

GMFCS Gross motor function classification scale

MACS Manual ability classification scale

CFCS Communication function classification scale

KIM Key informant methodology

LMIC Lower middle income country

8 Visual impairment and perceptual visual disorders in children with cerebral palsy in Nigeria

Authors

Roseline E Duke1,2 Kennedy Nwachukuw2, Chima Torty2, Uche Okorie2, Min J Kim1, Kathryn Burton4, Clare Gilbert1, Richard Bowman1

Affiliation

1London School of Hygiene &Tropical Medicine, International Centre for Eye Health, Keppel Street, London

2 University of Calabar Teaching Hospital, Children's Eye Centre, Department of Ophthalmology, Calabar, Cross River State, Nigeria.

3 University of Calabar Teaching Hospital, Department of Paediatrics, Calabar, Cross River State, Nigeria.

4 Cambridgeshire Community Services, UK

Corresponding authors contact

Email address: dr.roselineduke@gmail.com Phone: +234-8061302925

Key words: cerebral palsy, children, ocular visual impairment, cerebral visual impairment, perceptual visual disorders, neuro-ophthalmic,Nigeria

Synopsis

51% of children with cerebral palsy had ocular abnormalities, almost half had cerebral visual impairment. Visual dysfunction was associatedwith oculomotor dysfunction. Only

14% of carers recognized visual problems, highlighting the need for greater awareness.

1.11 ABSTRACT

Cerebral palsy (CP) is the most common cause of childhood physical disability globally. This study describes the spectrum of ocular morbidityand visual impairment in a community based (recruited by key informants) sample of children with CP in Cross River State, Nigeria.

Methods

A paediatric neurologist clinically confirmed CP and assessed systemic comorbidity. Ophthalmological assessment included developmental ageappropriate acuity tests, objective refraction, and objective and subjective tests of perceptual visual dysfunction (PVD).

Results

388 children aged 4 - 15 years with CP were identified. Visual problems were reported by carers in only 55 (14%) cases. Binocular visual acuityimpairment (BVAI) was seen in 20/201 by Lea symbols test (10%), and 213/388 (55%) by the mirror test. Abnormal visual fields were seen in 58/388 (14.9%); strabismus in 183 (47%) abnormal contrast sensitivity in 178 (46%) and abnormal saccades in 84 (22%), spherical refractive errors in 223(58%), significant astigmatism in 36 (12%), accommodative dysfunction in 41(10.6%), optic atrophy in 198 (51%). Perceptual visual disorders were present in 22 (6%) subjectively and 177 (46%) objectively.

The estimated frequency of cerebral visual impairment (CVI) in children ranged from 61(16%), to 191(49%) if children with optic atrophy wereincluded.

Conclusion

Children with CP have a wide spectrum of ocular morbidity and visual impairment, underestimated by carers. Children with CP require visual acuity assessments with a range of tests which account for associated comorbidities and oculomotor dysfunction. Functional vision assessmentsfor PVD is important. CVI is common.

1.12 INTRODUCTION

Cerebral palsy (CP) is the most common cause of motor dysfunction and one of the three most common lifelong developmental disabilities in children worldwide.(139) CP comprises a group of permanent and non-progressive disorders of movement and posture caused by a central nervous system lesion, damage or dysfunction originating early in life. This is accompanied by associated comorbidities.(27) The prevalence ofCP varies between 1.5 and 3.0 per 1000 live births, and is higher in low-resource settings.(29)

Ocular and visual problems are said to occur in 42.5% to 62% of children with CP.(192) The spectrum of ocular/visual problems in children withCP is broad, including refractive error, oculomotor abnormalities, optic atrophy and cerebral visual impairment (CVI). Detailed visual assessments from sub Saharan Africa are rare. CVI was recently defined in a systematic review, as “verifiable visual dysfunction not attributableto ocular pathology”,(84) although the best ways of assessing abnormalities in visual function in CVI are not universally agreed CVI can includea reduction in visual acuity and visual field defects as well as ‘higher’ visual perceptual problems or cognitive visual problems. (193) For the purpose of this study, problems with these higher visual functions will be termed perceptual visual dysfunction or PVD. PVD can be assessed objectively using a battery of tests,(194) and subjectively using clinical/behavioural question inventories for example, the Insight Inventory Questions (IQI).(96)

The aim of this study is to describe the full spectrum of ocular morbidity and visual impairment including PVD, and their relation to other

disability/co-morbidity in a population based sample of children with CP in Cross River State, Nigeria.

1.13 METHODS

1.13.1 Study design and participants

A community based sample of children with CP were recruited to participate in a clinical trial to assess the impact of child-specific interventionsarising out of the IQI.(163) Children were identified in three districts in 18 local government areas (LGA), Cross River State, Nigeria between December 2017 and September 2018.

1.13.2 Sampling

We estimated the population of children in the study area to be 1.1 million, and calculated the sample size assuming that a least 50% of childrenwith CP would have CVI,(60) using 5% precision and a 95% confidence level. The minimum sample size was 383.

1.13.3 Identifying cases, recruitment and case ascertainment

The key informant method was used to identify children suspected of having CP in the community lay key informants were identified by community leaders and underwent one day of training. A range of search methods were used, including house to house visits, school and churchvisits. Referred children were examined by a paediatric neurologist at a health centre and those aged 4-15 years with confirmed CP whose families consented were recruited. CP was defined according to history and neurological examination in line with international criteria.(28) Gross Motor Function Classification Scale (GMFCS) was used to describe the severity of motor impairments based on a child's current motor

abilities.(28) These levels were categorized into ambulatory (Levels I–III) and non-ambulatory (Levels IV–V).(170) The Manual Ability Classification System (MACS) and other outcome measures included the subtype of CP, classified by the Surveillance of Cerebral Palsy inEurope (SCPE), and an anatomic classification.(42)

1.13.4 Comorbidities

Comorbid conditions, which were confirmed by standardized clinical evaluations by a paediatric neurologist, included feeding and swallowingdifficulties and malnutrition, epilepsy, impairment of hearing, cognition, speech and learning, and communication difficulties (communicationfunction classification scale 4-5).(48, 175, 177, 182, 195) Psychological assessment (emotional and conduct problems, hyperactivity, peer problems and prosocial behaviors) were assessed using the strength and difficulties questionnaire (SDQ). The parent completed SDQ questionnaire was administered by trained medical social workers. Symptoms scores were analysed online (by youthinmind) using the originalthree-band categorization and 5 scales, thereafter generating the total difficulties score we refer to as abnormal behaviors. (179)

Detailed ophthalmological history and examination were conducted by a paediatric ophthalmic team, including an ophthalmic nurse, optometristand a paediatric ophthalmologist (Table 1).

In brief, assessment for abnormal accommodation, abnormal saccades, abnormal visual field defects are described below.

1.13.5 Assessment of accommodation

The near pupil responses technique was performed because the cognitive demand on the child is low as the child passively observes the target asit moves and is easier to perform. The examiner places his face while using a torch midline of the patient in a fixed position and distance (50cm)and the target (a yellow and red sticker at the top on a sick) is placed 33cm away from the child eyes. The child fixates on the target while the examiner looks at the pupillary reaction assessed subjectively by the retinoscope graded as (subjectively as normal, abnormal (reduced, or absent). A normal response was recorded if a brisk constriction was observed on near fixation, such as the examiner would expect from a child with normal vision.

A reduced response was recorded where limited or slow pupil constriction was observed. If there was no perceptible changein pupil state with near fixation the response was recorded as absent.

1.13.6 Assessment of saccades

In non-verbal children, two fixation targets were used. The child was enticed to look at the tester's face at about 50cm. When fixation is midline,one object is presented 20-30cm from the midline. The infant or child was again enticed to look at the other object presented on the opposite sideof the examiners face. For verbal children, the patient was asked to look at the examiner's nose and then at the examiner's finger to the left followed by the right of central fixation only upon verbal command.

Normal saccades: quick, simultaneous movement of both eyes between two or more phases of fixation in the same direction.

1.13.7 Assessment for visual field defects

Binocular visual field assessment was conducted. The examiner observed when the young child's eyes move in a quick saccade or (blinking the eyes, smiling, excitement) from the straight-ahead position or from fixation of the examiners face to the flickering stimulus of intensity 400 cd/m2 when measured at the side of the diode of the LEA Flicker Wand. The flexible wand was bent in a half circle. The examiner stood on the child's side so that the child's eyes could be seen. The LEA Flicker Wand was placed behind the child's head and the flickering stimulus broughtforward on the child's right side at 40 cm from the child's head. The flickering stimulus was brought from the back of the child's head up in the child's left and right upper field quadrant and below the childs face in the lower right and left quadrant. To estimate the size of the visual field, the point at which the child responded to the flicker in all four

quadrants was noted.

1.13.8 Visual acuity assessment and refraction

Chronological and developmental age appropriate tests for uncorrected visual acuity were performed using the Lea Symbol LogMar Test, themirror test (7) and an assessment of light fixation and following (for details see Table 8.1).

Levels of visual acuity assessed using Lea symbols were classified as follows: LogMar ≤0.8 – normal vision; >0.8 -1.0 low vision, >1.0 -3.0 blind. Lea grating detection scores in cycles per degree were converted into LogMAR; e.g., 4 cycles/sec= 0.9 and 8 cycles/sec =0.6. In the mirrortest, the distance at which a child fixates their reflection correlates with logMAR acuity assessed by preferential looking.(7) In this study we usedthe same conversion in which a mirror distance of 110cm was considered normal vision, 40-60cm as low vision and <40cm as blind.(7) The light fixation test was used to categorize children into: normal vision (fixing and following), low vision (fixing but not following) and blind (not fixing or following). Binocular visual acuity impairment (BVAI) was defined as low vison or blind visual acuity categories for analysis of risk factors.

Cycloplegic refraction was undertaken (details in Table 1) and the most ametropic meridian (MAM) was derived for each eye. Refractive errorswere sub-categorized as low, moderate, high.(196)

8.3. 9 Subjective assessments of PVD (Insight Questions Inventory)

PVD was assessed subjectively using the Insight Questions Inventory (IQI) which has 52 questions in seven sections (for details see Table1).(96) The IQI was translated into contemporary English and the local language and administered to each carer who identified

their observations of the visual difficulties experienced by their child. Each of the 52 questions has a 5-point Likert scale (0-5) to allow carers to describe whether their child has the problems never, rarely, sometimes, often or always. They could also indicate “not applicable”, for example,for questions relating to mobility if the child was not mobile.

Questions in the IQI in which more than half of the respondents reported not applicable were excluded from analysis. A child was arbitrarilydefined as having PVD if their mean score was ≥3 out of 5 for one or more of the 7 sections.

8.3.10 Objective assessment of PVD

Twelve objective tests for PVD were conducted, including the Lea 3-D puzzle (coloured and black and white), Lea mail box game (horizontal, vertical and oblique), Lea rectangles with grasp of the blocks, appreciation of speed, size and length, pickup test on plain and patterned surface, facial recognition test and facial expression interpretation.(194) The small numbers of missing variables were counted as passes based on clinicaljudgement (Supplementary material 1). Objective PVD was defined dichotomously as failure of more than 6 of the 12 tests.

8.3.11 Definitions of cerebral visual impairment

CVI was defined as: Group A) BVAI in the absence of ocular pathology such as significant refractive error, optic atrophy or other causes ofvisual loss, or visual field defects without an ocular cause in the presence of normal vision acuity. In Group B): the definition of CVI was

expanded, to include children who had optic atrophy as the only ocular abnormality or they have evidence of PVD but normal visual acuity withno ocular pathology.

8.3.12 Data analysis

All analyses were carried out using STATA 11. Descriptive statistics were reported using means and standard deviations (for continuous measures) or frequencies and percentages (for categorical variables). Comparisons between categorical variables were performed using the chi- square test. Proportions and frequencies were assessed for visual impairments, ocular signs, systemic comorbidities and CP severity and subtype.Paired t-tests were used for comparison of right and left eye refractive errors. Binocular visual acuity (BVA) was analysed categorically for two visual acuity tests (the mirror test and Lea symbols test) (Table 1). Multiple logistic regression models, which included age and sex, were developed to identify factors associated with BVAI assessed by the mirror and Lea tests and also for, objective PVD (multiple logistic regression) and subjective PVD (linear regression).

Variables included in the regression models for both BVAI and PVD (objective and subjective) included systemic comorbidities, clinical types ofCP and severity, ocular signs, and age and gender (variables as described above). These were chosen based on biological plausibility and findingsfrom previous studies. In addition, a no selection procedure was used to include other factors: variables significant at $p<0.2$ level, or not significantat $p>0.2$ but with an odds ratio between 0.5 and 2.0 in bivariate logistic regression were also included in the multivariate model.

8.3.13 Ethical approval

The study was performed in accordance with the Helsinki declaration and approved by the ethics committees of Cross River State and the London School of Hygiene & Tropical Medicine. The study was explained to carers in their preferred language, mostly in English. A writteninformation sheet was read out. Assent from older children was sought. Parents signed the consent if they agreed. No parent declined participation. Children were referred for surgery, were prescribed and dispensed glasses as needed.

1.14 RESULTS

1.14.1 Demographics

A total of 388 children with CP were enrolled. Their mean age was 9.2 ± 4.0 years and the median age was 9 (IQR 6 to 13) years. 159 (41.0%%)were female. The spastic form of CP was the most common (271, 70%) followed by ataxic (38, 10%), choreo-athetoid (29, 7%) dystonic (18,

5%), and unclassified (32, 8%). 280 (72%) were ambulatory (GMFCS I-III) and 300 (77%) had moderate or good manual dexterity (ManualAbility Classification Scale 1-3).

1.14.2 Comorbidities

The following comorbidities were identified: impairments in learning 342 (88%), cognition 269 (69%), hearing 46 (12%) and speech 331

(85%). Feeding and swallowing difficulties were identified in 334 (86%), and 299 (77%) children respectively, and 200 (51%) were

malnourished. Other comorbidities included communication difficulties 173 (45%), epilepsy 130 (35%) and abnormal behaviour 231 (62%) which include (reduced kind and helpful behaviour 252 (67 %), difficulties getting along with other children 240 (63%), abnormal conduct 233(62 %), hyperactivity and inattention 162 (43%).

1.14.3 Visual function

Poor vision was reported by 55/388 (14%) carers. Post-secondary education certification was obtained by 79(20%) fathers and 60 (15%) mothers. (Supplementary material 2). Quantitative preferential looking using Lea grating was achieved in 32/388 (8%) children, with 2/388 (1%)in the low vision category.

BVAI was identified in 231/388 (59%) children using the mirror test, 81/388 (21%) using light fixation test and 20/201 (10.0%) with Lea symbol test. 187/388 (48.2%) could not be assessed with Lea symbols for cognitive reasons (unable to recognize or match letters/symbols). Three children (1.5%) were in the worst category of visual acuity for all three tests and 94 (46.8%) were in the best category for all three tests

(Table 8.2). The mean age of children with normal vision with the mirror test was 9.3 (SD 4; IQR (6,13)), Lea symbols 10.1 (SD 4; IQR (7,14))and Light fixation 9.2 (SD 4; IQR (6,13)). (Supplementary material 3).

Binocular visual field loss was identified in 58/388 (15%) children; homonymous hemianopia in 8%, inferior in 3% and superior in 4% children.Almost half the children had abnormal contrast sensitivity (178/388, 46%), which was associated with difficulty in identifying faces 137/178 (77%) (OR 2.6, $p<0.001$) and in interpreting facial expressions 136/178 (76%) (OR 2.6, $p<0.001$).

1.14.4 Ocular abnormalities and neuro-ophthalmic signs

Lesions of the optic nerve predominated, generalized atrophy (34%), sectoral optic atrophy (9%) and a high cup: disc ratio (7%) (Table 4). Almost half of the children (47%) had strabismus, and constant exotropia was the most common type (Table 4), 22% had abnormal saccadic eyemovements.

1.14.5 Refractive error (RE)

No significant difference was seen between right and left eyes ($p=0.59$) and so data for right eyes are reported. Data on refractive status were obtained from cycloplegic auto refraction in 239/388 (62%) and cycloplegic retinoscopy in 111/346 (29%) children. The most ametropic

meridian (MAM) characterized refractive error which ranged between -17D and +15D; mean -0.14D, SD ± 2.3 (Figure 1). 165/388(42%) of children were emmetropic (refractive error in the range -0.50D to +1.0D). (Categories of refractive errors are defined in Table 8.1as low, moderate and high). MAM did not vary significantly with age (coef 0.2; p=0.38) and sex (coef=0.09; p=0.79). A multivariate analysisrevealed no significant association between MAM and CP type or ocular or systemic variables.

Abnormal accommodative responses were detected in 97/388 (25%) children; 23/97 (24%) were hypermetropes and it was more common (OR1.3, p=0.002 CI 1.0-1.9) in emmetropes 74/97 (76%).

108 children had spectacles dispensed; 58/108(54%) parents did not collect the spectacles while 50/108 (46%) parents collected spectacles butreported poor compliance (lack of constant wear) in 38/50 (76%) children.

1.14.6 Perceptual visual dysfunction

Eight 8/52(15%) questions in the IQI (Questions 9,5,15,32,33,34,45 and 46) were expunged from subjective PVD analysis because of high levelsof 'non-applicable' responses. Subjective PVD was identified in 22/388 (6%) children from IQI responses with a mean score of 3.04; SD2.43.

The proportion of children that passed the objective PVD tests are seen in supplementary material (Appendix 12.9). 177 (46%) children fulfilledthe criteria for objective PVD. 15 (4%) children met both sets of criteria for PVD.

1.14.7 Cerebral visual impairment

Forty-one children had BVAI (mirror test) without any ocular abnormality, and a further 20 had a visual field defect with normal acuity in theabsence of an ocular abnormality. This gives a minimum estimate of the proportion children with CVI of 61/388 (16%) (Group A)

(Table 5).

Using the expanded definition (Group B), the following children can be included; those with isolated PVD (27); and children with BVAI andoptic atrophy as the only ocular abnormality (103). This increased the number to 191 (49%).

1.14.8 Factors associated with binocular visual acuity impairment

In age and sex adjusted multivariable analysis, BVAI was not associated with the type or severity of CP or the presence of comorbidities, but was associated with abnormal saccadic eye movements ($p \leq 0.01$), objective measures of PVD ($p \leq 0.001$) and cylindrical refractive errors ($p \leq 0.05$) (Table 8. 3).

1.14.9 Factors associated with perceptual visual disorder

In age and sex adjusted multivariable analysis, objective perceptual visual disorder was significantly associated with BVAI (Mirror test) (OR 0.5; p=0.010, CI 0.29-0.85), abnormal contrast (OR 0.3; p<0.001, CI 0.20-0.59), abnormal saccades (OR 4.2; p<0.001, CI 1.8-10.0), abnormal behaviours (OR 2.2; p=0.003, CI 1.26-3.57), cognitive impairment (OR 2.4; p=0.03, CI 1.09-5.24), choreoathetoid CP (OR 5.7 p=0.012; CI 1.5-22.1). Table 8.3.
Abnormal saccadic eye movement was the only variable significantly associated with both measures of PVD and with BVAI using the mirrortest.

1.15 DISCUSSION

A wide range of complex visual and ocular pathology was found and was under-recognized by carers. Despite 59% of children having BVAI based on the mirror test, only 14% of parents reported visual problems. The rates of BVAI varied significantly according to how acuity was tested eg recognition/matching tests showed lower rates but were performed successfully only by the more cognitively able children who are lesslikely to have severe brain injury.

Despite visual acuity tests being conducted by an experienced paediatric optometrist, very few were able to give a reliable response to the grating preferential looking test which has been reported in other severely affected groups. (197) In contrast, using a mirror to elicit fixation ofthe child's own face was a useful and discriminatory test in this group of children, perhaps because the face is a stronger stimulus, and this testhas been shown to correlate quantitatively with preferential looking acuities in a group of normally developing children.(7)

Optic atrophy was the leading ophthalmological finding in our study, similar to other studies.(198) In CP it is thought to reflect anterograde degeneration of the retinal ganglion cells rather than a separate insult.(74) Oculomotor abnormalities were common, as almost half had strabismus and almost a quarter had abnormal saccades and smooth pursuit. Abnormal saccades were associated with both BVAI and PVD, suggesting that cerebral visual impairment can manifest itself through oculomotor dysfunctions, such as impaired fixation, smooth pursuit, andsaccadic movements.(193) Exotropia was more common than esotropia in contrast to previous studies of CP.(199) This may represent ethnic

differences, as exotropia is more common than esotropia in African children, but may also reflect differences in the type of brain injury causing the CP in a population where birth

asphyxia may be a more common cause than prematurity.(200, 201) Strabismus surgery should be consideredin children with CP for psychosocial reasons as well to enable ocular realignment and binocular vision.(199, 200)

At least 16% of children in our study had CVI, using BVAI and visual field abnormalities in normal appearing eyes as the criteria. However,optic atrophy is common as an epi-phenomenon in CVI; allowance for this and for PVD with intact acuity (rare in this population) gave an expanded estimate of 49%.

Reduced accommodative responses, which occurred in 25% of our population, can impair near and distant (in hypermetropes) vision, and can lead to amblyopia.(202) Good near vision is also important for visual development. The link could also be in the opposite causal direction in that children whose acuity is poor may have less drive to accommodate even though the efferent pathway is intact. This finding highlights the need toassess and correct poor accommodation in this group of children.

In our study, subjective PVD scores were consistently lower than reported in other populations.(91, 92, 203) This may be because children in ourstudy had more severe CP and a less integrated environment, making questions relating to acts of daily living less relevant, such as those relatingto mobility, for example. However, greater severity of CP is unlikely to be the whole explanation as 72% of children were ambulatory. Another

explanation is a lower level of interaction of parents with their children than in some other cultures, making parents less confident about answering functional questionnaires. In addition, cultural beliefs are likely to play a very important role in our society where disease / disability is stigmatizing and believed to have origins in spirituality.(51) In contrast to the subjective

assessment, objective assessment of PVD identified ahigher proportion of children with PVD and maybe more useful in this setting.

There is debate over whether children with CP and normal visual acuity but clear evidence of PVD should also be classified as a type of CVI. However, in this population, very few children had isolated PVD without BVAI or visual field defects. The reason for assessing PVD is not, therefore for screening, but to gain useful information about specific functional visual problems to aid habilitation and possibly a better quality of life. Previous work has shown that subjective PVD influences quality of life in children with CP independent of acuity impairment and that improvements in quality of life may be possible through understanding and adapting to PVD, suggesting that a broader definition of CVI whichinclude PVDs is appropriate.(91, 92)

A weakness of our study was lack of corrected visual acuity data, but spherical refractive error did not seem to be a direct cause of visual impairment in this study. The proportion of children with spherical error and significant cylindrical refractive errors was lower than in other studies, although most other studies are hospital based which may reflect selection bias. (204, 205). Despite counselling, only just over half theparents collected their child's spectacles and only a quarter subsequently wore them. Furthers studies are required on barriers to the uptake and

use of spectacles and the impact of correction on visual and general development in children with CP. The tests for saccades, pursuits and accommodation were clinical and limited in some cases by visual impairment and also subject to intra and inter-observer error and, hence theremay be measurement bias.

In conclusion, children with CP need to have a comprehensive systemic and visual assessment because significant visual impairment and morbidity is common and under-recognized by carers. Better assessment of children and education of carers may help to accurately defineappropriate and available interventions and improve the child's quality of life and opportunities for social integration and education.(193)

Funding: The Queen Elizabeth Diamond Jubilee Trust/Common Wealth Eye Health Consortium funded the research. Grant number [LSHTMITCRZ6814].

Acknowledgments: We acknowledge all the carers of children with CP and the key informants who participated in this study.

Contributorship: Conception and design of the work: Richard Bowman, Roseline E Duke, Kathryn Burton; Data collection: Roseline E Duke,Kennedy Nwachukuw, Chima Torty, Uche Okorie; Data analysis and interpretation: Roseline E Duke, Min J Kim; Drafting the

article:

Roseline E Duke; Critical revision of the article: Kathryn Burton, Clare Gilbert, Richard Bowman; Final approval of the version to be published and agreement to be

accountable for all aspects of the work: Roseline E Duke, Kennedy Nwachukuw, Chima Torty, UcheOkorie, Min J Kim, Kathryn Burton, Clare Gilbert, Richard Bowman.

Competing interests: The authors declare that they have no competing interests.

Table 8.1: Ophthalmic assessment of visual function

Visual function	Test	Tester
Presenting distance visual acuity	Lea symbols - The Lea symbol test was conducted by a trained optometrist, using an illuminated Lea symbols LogMar chart and symbol cards. The examination distance was 3 metres and a 3/4 criterion was prescribed.(206)	Optometrist
Mirror test	The test was conducted by the Pediatric Ophthalmologist on all children. A child was positioned approximately 20 cm from a mirror until they were deemed to attend their own reflection. The child was then moved slowly back until fixation was lost, as evidenced by the head and eyes turning away. Three distances were assessed, to represent blind, low vision and normal respectively - <20cm, 20cm-30 cm and 550cm. (Because of reflection, the actual viewing distance is twice the distance from the child to the mirror -	Pediatric Ophthalmologist
	40cm, 60cm, 110cm.) The mirror-to-child distance was measured from an already marked floor using a tape measure. The process was carried out once. Hand-held mirror of size of 30 cm by 40 cm was used.(7)	

Light fixation and follow test	Both eyes assessed binocularly. A penlight source was used to assess the ability to fix and follow. • Inability to maintain fixation or follow =blind. • Fixation alone = low vision • Fixation and following = normal vision.	Paediatric Ophthalmologist
Grating detection test (preferential looking PL)	Lea grating test which uses paddles to present grating was used to assess grating detection in children in which Lea symbols test was below the normal category, those with perception of light (PL) vision and in uncooperative children. Children were assessed binocularly.	Optometrist
	Lea gratings used, measured 0.25 cpcm, 1 cpcm, 2.0cpcm, 4.0cpcm and 8 cpcm gratings were used at 57 cm. The grating was kept behind the grey surface while moving it in the midline to the testing distance. Then the grating and the grey surface are moved in opposite directions. Measurement was based on observing the child's eye movements when the grating paddles were presented. The test was stopped if the child showed no interest, looked confused and did not follow the movement of the grating. (194)	

Contrast sensitivity	Hiding Heidi Low Contrast 'Face' under photopic condition(194). Single Sided: Seven cards printed on one side in the following contrast levels: black, 25%, 10%, 5%, 2.5%, and 1.25%. Contrast sensitivity was described as not assessable-abnormal or assessable-(normal ≥2.5%)	Optometrist
Colour vision	Lea colour vison screening plates. Categorized as no colour problems or abnormal colour vision (red/green blindness or weakness, total colour blindness or yellow blue defects)	Optometrist
Visual fields	Lea Flicker Wand 280000 was used to assess major field losses.	Pediatric Ophthalmologist
Eye movements		Pediatric Ophthalmologist
Vergence	Observation of binocular movement of the eyes i.e., convergence and divergence, using a fixation target.	Pediatric Ophthalmologist
Smooth pursuit (horizontal and vertical)	• Non-verbal, uncooperative or inattentive children: slowly rotate a mirror (10 x 15cm) held before their eyes. • Verbal children: the patient is asked to track the tip of a blue ball point pen held a meter before the eyes with the head still. The target is moved at a low, uniform speed.	Pediatric Ophthalmologist

	Normal smooth pursuit movements: eyes move smoothly instead of in jumps and abnormal (absent, discontinuous)	
Saccades	• Non-verbal: two fixation targets are used. The child is enticed to look at the tester's face. When fixation is midline, one object is presented 20-30cm from the midline. The child is again enticed to look at the tester's face after which the other object is presented on the other side. • Verbal: The child is asked to look at the examiner's nose and then at the examiner's finger to the left or right of central fixation only upon verbal command. Normal saccades: quick, simultaneous movement of both eyes between two or more phases of fixation in the same direction.	Paediatric Ophthalmologist
Strabismus	Corneal reflex test and Modified Krimsky test.Defined as esotropia, exotropia, hypotropia or hypertropia	Paediatric Ophthalmologist
Accommodation	Near Pupillary Response technique assessed with the use of an accommodative target which provided an indicator of accommodative pupillary response in children as described by Saunders.(207)	Pediatric Ophthalmologist
Cycloplegic refraction	Cyclopentolate 2% + Phenylephrine 2.5%; Zeiss autorefractor/ keratometer; manual objective retinoscopy in those not cooperative for autorefraction. The most	Optometrist Paediatric

	ametropic meridian (MAM) were derived for each eye and refractive error categorized as shown below. (208) • Emmetropia: MAM> -0.75 to +1.00 D • Low to moderate hypermetropia: MAM > +1.00 to +4.00 D • High hypermetropia: MAM > + 4.00 D • Low to moderate myopia: MAM - 4.00 to > -0.50 D • High myopia: MAM >- 4.00 D	Ophthalmologist

	Significant astigmatism was defined as astigmatism ≥ 1.00 DC Criteria for dispensing glasses: myopia≥ 2D, hypermetropia ≥ 4D, astigmatic cylinder ≥ 2D, in accordance with the Cross River state eye care programme dispensing guideline for children.	
Anterior segment examination	Detailed anterior segment examination was performed using a portable slit lamp and where not possible, a pen torch with magnifier was used.	Paediatric Ophthalmologist
Posterior segment examination	Dilated fundus examination using a binocular indirect ophthalmoscope with 28D or 20D lenses. In addition to direct fundoscopy to assess the optic disc, fundus photos were taken using the Horus 2010 fundus camera. Optic neuropathy was defined by a) the number of quadrants with optic disc pallor (sectorial (horizontal /vertical bands), generalized b) enlarged cup (axial), and c)	Paediatric Ophthalmologist

	reduced optic disc diameter (hypoplastic).	
Structured questions inventory to identify subjective PVD (Insight Questions Inventory)	The 52 questions are in seven sections; each questions has a 5-point Likert scale to allow carers to describe whether their child had the problems never =1, rarely =2, sometimes =3, often =4 or always= 5. They could also indicate "not applicable" =0. Each section covers different visual perceptual abilities: section 1 has 11 questions on visual fields; section 2 on perception of movement (5 questions); section 3 - visual guidance of movement (7 questions), section 4 - searching for visual targets (12 questions); section 5 - noticing multiple targets and visual attention (5 questions), section 6 - recognizing target objects and also navigating around (11 questions) and section 7 - visualizing crowded scenes (5 questions).	

Table 8.2: Levels of visual acuity assessed using four different visual acuity tests in children with cerebral palsy

Visual acuity test (proportion assessed)	Details	N	%
Lea symbol test (51.8%)	Normal vision: LogMar <= 0.8	181	46.6
	Low vision: LogMar >0.8-1.0	9	2.3
	Blind: LogMar 1.0-2.5	11	2.8
	Unable to do test: LogMar 3	187	48.2
Mirror test (100%)	Normal: attentive at 110cm	175	45.1
	Low: attentive at 40-60cm	103	26.5
	Blind: attentive at <40cm/non attentive	110	28.4
	Unable to do test	0	0
Light fixation test (100%)	Normal: fixes and follows	307	79.1
	Low: fixates only	30	7.7
	Blind: no fixation, no following	51	13.2
	Unable to do test	0	0
Grating/Preferential looking (8.2%)	Normal < 8 cycles/sec	30	7.7
	Low: 4 cycles/sec	2	0.5
In BVAI category for all three tests		3	1.5
In normal category for all three tests		94	46.8
TOTAL		388	100%

BVAI (mirror test) = 213/388 (55%)

Table 8.3: Factors associated with binocular visual acuity impairment using mirror and Lea symbol tests and objective and subjective perceptualvisual disorder test in an age and sex adjusted multivariate logistic and multiple linear regression

	Binocular visual acuity impairment		Perceptual vision disorder	
Variables	Mirror Test (213/388)	Lea symbol Test (20/201)	Objective PVD (177/388)	Subjective PVD (22/388)
	Adjusted odds ratio (95%CI)	Adjusted odds ratio (95%CI)	Adjusted odds ratio (95% CI)	Adjusted coefficient (95% CI)
Sociodemographic				
Age group >9 years	1.0	6.1	1.4	0.9
Sex (male)	0.8	7.7	0.8	0.06
Visual function / PVD				
Abnormal saccades	**0.3 (0.15-0.83)	0.02	***4.2 (1.8-10)	*0.3 (0.10-0.50)
Objective PVD	***0.5 (0.28-0.82)	2.5	NS	NS
BVAI (mirror test)	NS	NS	*0.5 (0.29-0.85)	0.001

Abnormal contrast sensitivity	0.8	2.0	***0.3 (0.20-0.59)	-0.1
Ocular abnormalities				
Cylindrical refractive errors	**0.3 (0.11-0.68)	1.2	NS	*0.2 (0.004-0.43)
Strabismus	0.8	9.7	0.8	0.04
Spherical refractive error	NS	3.5	0.9	NS
Abnormal smooth pursuit	0.5	6.8	0.5	-0.1
Abnormal accommodation	1.5	0.003	0.8	0.05
Nystagmus	NS	NS	2.7	0.2
Myopia	NS	NS	1.5	0.1
Optic atrophy	NS	NS	NS	0.1
Type and severity of cerebral palsy				
Spastic	NS	9.7	1.8	NS
Choreoathetoid	0.4	NS	*5.7 (1.5-22.1)	NS
Tetraplegia	1.5	NS	NS	NS
Communication Function Classification scale 4-5	0.6	0.03	1	0.02
GMCS score 4-5	1.1	9.8	NS	-0.1
MACS score 4-5	0.8	7.4	NS	0.2
Comorbidities				
Feeding difficulty	2	1.7	NS	NS
Cognitive impairment	0.8	2.1	*2.4 (1.09-5.24)	0.002
Learning impairment	NS	NS	1.8	NS

Epilepsy	0.8	0.8	1.1	0.1
Swallowing impairment	1.1	17.8	-0.1	-0.1
Speech Impairment	0.8	8.2	0.6	0.1
Hearing impairment	0.9	4.9	1.3	0.1
Abnormal behaviours	0.9	NS	*2.1 (1.26-3.57)	*0.13 (0.01-0.26)
Malnutrition	0.7	96.0	NS	NS

*p<=0.05, **p<=0.01, ***p<=0.001

NS: Not significant in univariate

analysis and not included in multivariate

analysis GMCS = Gross Motor

Classification Scale; MACS = Manual

Ability Classification Scale

Table 8.4: Causes of ocular morbidity in children with cerebral palsy, by anatomical site and neuro-ophthalmic signs (n=388)

Anatomic site	Type of pathology	N	%
Optic disc	Generalized (vertical / horizontal) optic atrophy	134	34.5
	Sectoral (horizontal) optic atrophy	34	8.8
	Axial (high cup disc ratio)	29	7.5
	Congenital optic disc pit	2	0.5
	Chronic papilledema	1	0.3
Pupil	Abnormal pupil morphology	6	1.5
Retina	Hyper pigmentary changes in retina	22	5.7
	Retinal pigment atrophy	9	2.3

	Reduced retinal vascular tortuosity	2	0.5
	Increased retinal vascular tortuosity	1	0.3
Cornea	Cornea scar	6	1.5
Whole globe	Microphthalmos	5	1.2
	Megalocornea	1	0.3
Lens	Cataract	4	1
	Pseudophakia	1	0.3
Other	Albinism	2	0.5
Neuro-ophthalmic signs		N	%
Strabismus		183	47.2
Exotropia		130	71.8
Constant		118	30.4
Intermittent		10	2.6
Sensory		2	0.5
Esotropia		51	28.2
Infantile		40	12.7
Sensory		2	0.5
Hypotropia		1	0.3
Hypertropia		1	0.3
Abnormal saccadic movements		84	21.6
Abnormal smooth pursuits		71	18.3
Nystagmus		16	4.1
Abnormal pupil reaction		22	5.7

Table 8.5: Cerebral visual impairment in children with cerebral palsy with and without binocular visual acuity impairment assessed using themirror test (n=388)

Variables	Binocular visual acuity impairment 213 (55%)			Normal visual acuity 175 (45%)			Total
	Ocular abnormality			Ocular abnormality			
	Absent	Present	Subtotal	Absent	Present	Subtotal	
Total Numbers	N=41	N=172	N=213	N=81	N=94	N=175	N=388
Visual field abnormality	21	14	35	20	3	23	58
Objective perceptual visual disorder	20	104	124	27	26	53	177

Colour key

BOLD= definite CVI

Italics = includes the expanded definition of CVI

Supplementary material (Table 8.1): Ophthalmic tests for cerebral visual impairment/perceptual visual disorders

Subcategory of PVD	Test(194, 206, 209)	Observation	Grading (easy-pass:cannot and others- fail; Yes-pass, No-fail)
Visual guidance, colour, eye hand coordination and visual memory	1. Lea 3-D puzzle coloured	• Eye-hand coordination and visual guidance of movement: watching the movement a child makeswith their hand, fingers, elbow or forearm when asked to turn over a piece of puzzle. The capacity to orientate the shape is subjectively viewed. • Easy: places the puzzle pieces correctly	Easy, difficult, cannot do and why,or not applicable
Visual guidance and 3D recogn	2. Lea 3-D puzzle black & white	Eye-hand coordination and visual guidance of movement: watching the movement a child	Easy, difficult, cannot do and why,or not applicable

ition of concre te object s		makeswith their hand, finger elbow or forearm when asked to turn over a piece of puzzle.	

		Easy: can place the puzzle pieces correctly	
		Short-term memory for localisation: can a childplace a piece of puzzle in the correct place after being distracted	
Visual recogni tion and line orientat ion in three dimensi ons (vertica l, horizon	Lea mailbox game 3. Vertical 4. Horizontal 5. Oblique	Visual perception of line orientation done with thefingers, hand, or forearm: • Child is asked to match the orientation of the slot • Child is asked to drop a card through the slot ofthe LEA Mailbox Game Easy= can do this in different orientations without	Easy, difficult, cannot do and why,or not applicable

tal, oblique) & eye hand coordination		hitting the mail box	
Visual recognition of differences in size, length and direction of lines	Lea rectangles 6. Placement of blocks	• Watch the orientation of the hand and fingers as the child grasps a block • Can the child place a block of the same length ontop of another? Easy = can do this	Easy, difficult, cannot do and why,or not applicable

	7. Appreciation of short and long	Can the child appreciate short and long as a changein length? If yes, this is defined as a pass.	Yes, No, not sure, donot know, not applicable

	8.Grasp speed	Watch the speed of the hand and fingers as the childreaches to grasps a block Can the child reach for the blocks smoothly at a normal speed? If yes, this is defined as pass.	Smooth and normal reach, slow, fast, notapplicable
Visual search	9. Pick-up test: Picks up a small ball of uniform size (1mm) placedon a plain surface	If a child identifies and picks up the ball by anymeans it is defined as easy	Easy, difficult, cannot do and why,or not applicable
	10. Pick-up test: Picks up a small ball of uniform size (1mm) placed on a patterned surface	Identifies the ball and picks the ball by any means itis defined as easy	Easy, difficult, cannot do and why, or not applicable
Heide expressi on test	11. Facial expression interpretation	Can the child interpret the facial expression of the smiling Heidi cards and the weeping Heidi cards?	Easy, difficult, cannot do and why,

		The child is asked to choose a smiling or weeping card. If child is deaf, the child is asked to choose and match the type of face with from a	or not applicable

		selection ofcards. Easy = ability to identify both weeping and smiling faces	
	12. Facial recognition Test	Can the child match and identify a minimum set of 4 similar faces by recognizing the differences in thefaces on the cards? Easy = can match and spot the differences in at least 4 out of 6	Easy, difficult, cannot do and why,or not applicable

Supplementary material (Table 8.2): Distribution of Refractive errors in children withCerebral Palsy

Type of error	Sub type of refractive error	N	%
Spherical error	Emmetropia	165	42.3
	Hypermetropia		
	Low to moderate (MAM >+1.00 to +4.00D)	86	22.2
	High (MAM >+4.00D)	4	1.0
	Myopia		
	Low to moderate (MAM -4.00 to >-0.50)	122	31.4
	High (MAM >-4.00D)	11	2.8

Cylindrical error (astigmatism)	Significant astigmatism (>/= 1.00DC)	36	9.3
	Not significant <1.00DC	32	83.5
	No cylindrical error	28	7.2
Aphakia		0	0
Pseudophakia		1	0.3

Supplementary material (Table 8.3): Factors associated with objective PVD in an adjusted forsex and age multivariate logistic regression, and subjective PVD in a linear regression n=388

Variables for	Objective PVD Adjusted odds Ratio (95% CI)	Subjective PVD Adjusted Coefficient (95% CI)
Age > 9years	1.4	0.1
Sex (Male)	0.8	NS
Abnormal behaviour	*2.2 (1.29-3.63)	*0.13 (0.01-0.26)
Choreoatheoid	*5.7 (1.5-22.1)	NS
BVAI (Mirror Test)	*0.5(0.29-0.85)	0.001
Abnormal contrast	***0.3 (0.20-0.59)	NS
Abnormal saccades	***4.2 (1.8-10)	*0.3 (0.10-0.50)
Abnormal smooth pursuit	0.5	-0.1
Nystagmus	2.7	0.2
Strabismus	0.8	0.05
Spherical refractive error	0.9	NS

Myopia	1.5	0.1
Cylindrical refractive error	NS	*0.2 (0.004-0.440)
Optic Atrophy	NS	0.1
Unclassified CP	0.9	NS
Dyskinetic CP	0.6	-0.2
Spastic CP	1.8	NS
Epilepsy	1.1	0.1
Speech Impairment	0.6	0.1
Hearing Impairment	1.3	0.1
Learning Impairment	1.8	0.05
Cognitive Impairment	*2.4 (1.09-5.24)	0.002
Communication Function Classification 4-5	1.0	0.02
Swallowing Impairment	NS	-0.1
Gross Motor Classification Scale	NS	-0.1
Manual Ability Classification Scale	NS	0.2

*P<=0.05, **P<=0.01, ***p<=0.001

NS: Non-significant in univariate and not included in multivariate analysis

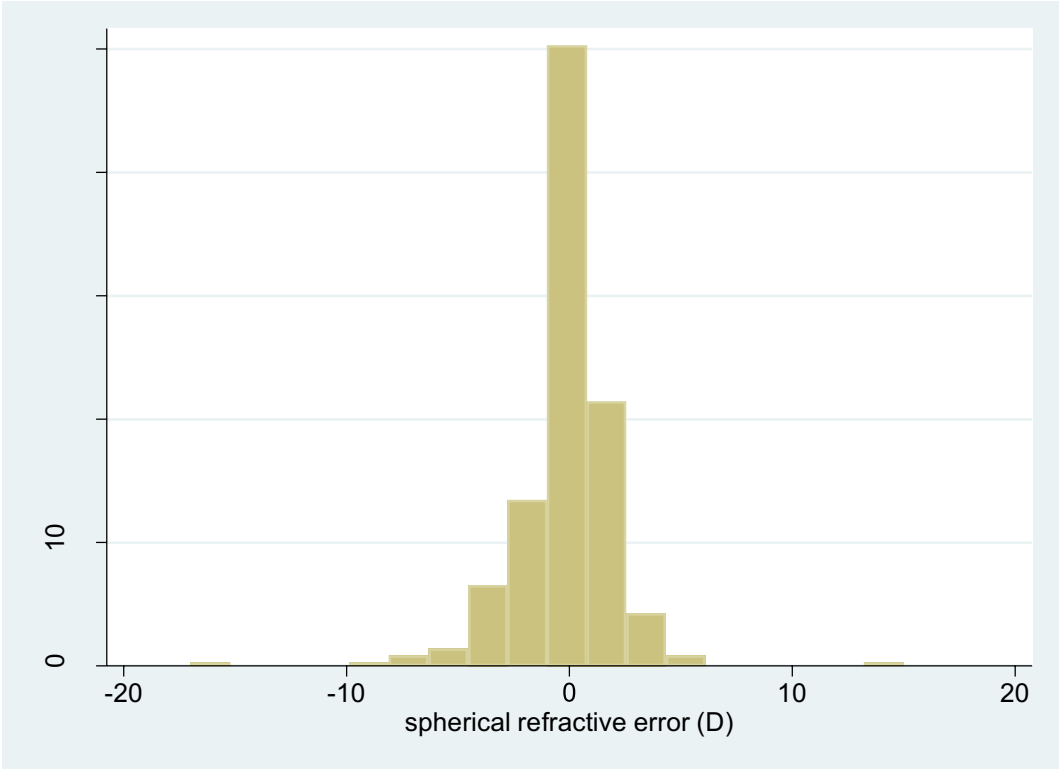


Figure 8.1. Distribution of spherical refractive error in children with CP

9 The effect of Insight Questions Inventory and Visual Support Strategies on carer-reported quality of life for children with cerebral palsy and perceptual visual dysfunction in Nigeria: a randomized controlled trial

Authors

Roseline E Duke*[1,2], Torty Chimaeze[3], Min J Kim[1], Soter Ameh[4], Kathryn Burton[4] Richard Bowman[1]

Affiliation

[1]London School of Hygiene and Tropical Medicine, International Centre for Eye Health,[2] University of Calabar Teaching Hospital, Children's Eye Centre, Department of Ophthalmology, Calabar, Cross River State, Nigeria.

[2]University of Calabar Teaching Hospital, Department of Pediatrics, Pediatric neurology, Calabar, Cross River State, Nigeria.

[4]University of Calabar Teaching Hospital, Department of Community Medicine, Calabar,Cross River State, Nigeria.

[4]Cambridgeshire Community Services, Oxfordshire

Corresponding Author:

*Dr Roseline E Duke

University of Calabar Teaching Hospital

Children's Eye Centre, Department of OphthalmologyCalabar, Cross River State, Nigeria

Email: dr.roselineduke@gmail.com

Keywords: Cerebral palsy, Cerebral visual impairment, Perceptual visual dysfunction, Insight Questions Inventory, Visual support strategies, Paediatric quality of life

Trial registration number: Pan African Clinical Trials

Registration, PACTR201612001886396. Registered on 3 December 2016.

Abstract

Structured clinical history question inventories have previously been used to try and elicit symptoms of perceptual visual dysfunction (PVD) in children with cerebral palsy (CP) in different settings. Earlier studies have suggested that PVD may affect quality of life and specific habilitational strategies, linked to inventory responses, may improve quality of life.Through an RCT, based on a community based sample of children with CP in Cross River State, Nigeria, we aimed to determine if a structured history inventory such as the Insight question inventory (IQI) and associated tailored visual support strategies (IQI VSS) for the management of those children who have PVD, can improve quality of life and is superior tostandard therapy.

Children with CP were recruited by the key informant method and confirmed by clinical examination. The parent reported IQI was used to identify children with PVD. Primary outcome measures were both Pediatric Quality of Life 4.0 Generic (PedsQL 4.0 Generic) andPediatric Quality of Life 3.0 Cerebral Palsy (PedsQL 3.0 CP) scale scores. Children were enrolled with a parallel arm allocation to either IQI and IQI VSS or to standard therapy for CP. Children were followed up for 6 weeks with weekly phone call session and the questionnaires repeated at the end of the 6 weeks' period.

Results show that the children in the treatment group (n = 191) showed no significantly different change between baseline and follow up in quality of life (PedsQL 4.0 Generic p=0.943: and PedsQL-CP 3.0 p=0.287), compared to the control group. There was suggestionof a better improvement (p=0.035) in the PedsQL 3.0 CP subscale of speech and communication for the intervention group.

The use of IQI VSS for the treatment of PVD in children with CP in this population does not show any superiority over current standard CP management in terms of overall quality of life. However, there was some evidence of improvement in quality of life in the area of speech and communication. Further research and refinement of these management method is required.

Introduction

Cerebral palsy (CP), is the most common neurologic and motor disability in children globally (210). CP describes a group of permanent disorders of the development of movement and posture causing activity limitation, which are attributed to non-progressive disturbances that occurred in the developing foetal or infant brain (28). The motor deficits are often accompanied by disturbances of sensation, perception, cognition, communication, behaviour,epilepsy, secondary musculoskeletal problems and nutrition (28).

World Health Organization, through the International Classification of Functioning, Disability and Health (ICF), clarified the understanding of CP in relation to intervention options and differentiated functioning problems, participation problems and disability. It issuggested that interventions should aim at maximizing a child's independence in daily activities and community participation, while also focusing on optimizing children's environment. In addition, a goal-based approach and (66), patient centeredness based on choice of interventions guided by what would best help the family achieve their goals, is recommended (93).

Visual impairment in children with CP varies in nature and severity and its prevalence ranges from 40-50% of children in different studies (60). Visual impairment in CP is often directly associated with the same brain injury which causes the motor problems. This is generally termed cerebral visual impairment (CVI) (84), commonly affecting children with CP but alsochildren with other neurodevelopmental diagnoses such as epilepsy and hydrocephalus. CVI can cause problems with 'basic' vision such as visual acuity or visual field or affect 'higher' visual perception or cognitive vision such as ability to see moving targets, to pick out a targetof interest from a complex scene, visual control of body movement or object recognition (90). This latter group of behavioural symptoms of abnormal higher-order visual processingcan

be termed perceptual visual dysfunction (PVD), part of the CVI spectrum.

It was previously shown in India, Bangladesh and more recently in the United Kingdom(91,92, 94), that this latter group of symptoms of higher visual processing problems or PVD are commonly present in children with CP; we have previously published a detailed visual assessment of the children recruited for this trial showing high rates of visual pathology (including 46% PVD measured objectively and 49% CVI (dropping to 16% if optic atrophy excluded) (95).

Previous work has suggested that PVD can be effectively assessed by a structured clinical history question inventories, including the insight question inventory (IQI) (91, 92, 96, 97). IQI scores have been shown to have internal reliability, and to discriminate between children diagnosed with CVI and healthy aged matched volunteers (68, 96) ; they have also been shown to correlate with neuropsychological tests of visual perception(92), and predict qualityof life in children with CP independent of other predictors such as visual acuity and degree ofmotor impairment (91).

The IQI provides in-depth information about the aspects of daily living activities that children struggle with. The current 52 item inventory, Insight Questions Inventory tests 6 domains of vision namely, visual field, perception of movement, visual guidance of movement, visual search, visual attention, recognition and navigation, which, in addition to visual field, test visual perception, either dorsal (occipito-parietal) stream processing (visuo-motor control, processing moving targets and processing large amounts of visual information at once) or ventral (occipito-temporal) stream processing (person and object recognition) (3).

Previous work indicated that considering dorsal and ventral stream as 2 factors explained 63% of variance of IQI scores between patients (94). In addition to diagnostic information, asimple software program links each question inventory response to a specific group of tailored visual support strategies (VSS) appropriate to that question, so that after completingthe inventory each

child/family has a set of tailored visual support strategies (IQI VSS) for that particular child. An example of the IQI and Visual support strategy can be seen in question 11, which asks “Does your child bump into door frames or partly open doors (left/right/both)”? With corresponding tailored visual support strategy response from caregivers who would give extra hints. For example, "There is a door coming up in a few steps." Another recommendation would be to replace doors with a beaded curtain.

A recent hospital based longitudinal study investigated the impact of the IQI VSS linked to the inventory responses on functional vision and quality of life (92). Children were followed up 6 months after receiving the IQI VSS and improvements were seen in both qualities of lifeand functional vision compared with baseline pre-intervention assessments but there was no control group.

We aimed to test whether this approach would be effective for a community based sample of children with CP in Cross River State, Nigeria by means of a randomized controlled trial. Wedid not screen for CVI or PVD within the CP sample but recruited all children with CP. The rationale for this was that we predicted high rates of PVD detectable with IQI and that almostall children with CP would have at least one positive response to IQI and therefore receive at least one IQI VSS.

Our previous work has suggested that PVD (measured by IQI) adversely affects quality of life,(91) and that IQI VSS improve quality of life (92), using the PedsQL 4.0 Generic module which assesses domains of physical activities, social, emotional and school functions. The PedsQL 3.0 CP module, is designed to be used by children with CP to detect changes arising from this condition or factors associated with it, with subdomains which include daily activities, movement and balance, fatigue, pain, school functions, eating and speech (23, 98, 99). Peds QL 4.0 Generic and PedsQL 3.0 CP assessments can be based on parent report, child reports or

proxy reports (25, 100). SincePedsQL3.0 CP is specifically designed for children with CP but has not been previously tested in relation to CVI or PVD we decided to use both PedsQL 4.0 Generic and PedsQL 3.0 CP modules as primary outcomes for this trial.

Methods

This was a parallel group, double blind clinical trial, with a superiority design. Recruitment took place between December 2016 to December 2018. Details of the trial methodology havebeen published and are briefly summarized here (163).

Participants

This prospective population study was conducted in 18 local government areas in Cross River State, Nigeria. Recruitment took place over 12 months, using the key informant method (36). In the first stage, a population based sample of children suspected to have CP were identified by the key informants with the use of the Ten questions Questionnaire and CP picture chart (169). In the second stage, children had a comprehensive history taken and detailed examination, including neurological examination and confirmation of the diagnosis and classification of CP by a paediatric neurologist who used the diagnostic criteria for CP(42).

Detailed visual assessment was performed and the results have been published including the Insight question inventory for PVD (95).

Eligible children were those confirmed to have CP who consented to be recruited into the trial (28). Data on all eligible children address and phone number(s) of carers was entered into a password protected database. Each child was allocated a unique identification number.

Assessment of CVI/PVD

For the purpose of the trial, symptoms of PVD were behaviourally ascertained by the use of the Nigerian Version IQI, a 52-item symptoms based inventory, (supplementary material 1), derived through linguistic translation of the British version of the insight questions inventory,and which was administered to each carer. There are 6 sections. Responses to each question was in accord with a 5-point Likert scale (1-5) to describe whether a child has problems: never, rarely, sometimes, often or always including "not applicable" respectively. The questions in each section are designed to identify CVI/PVD through asking about visual tasksinvolving both the dorsal (sections 1-5) and ventral (section 6) visual streams. Any subject who answered "sometimes", "often" or "always" to at least one question (out of 52) would beconsidered to have PVD and would receive at least one strategy. Questions with more than half of respondents reporting "n/a" were excluded (91).

Assessment of speech and communication

The Communication Function Classification Scale (CFCS) assessed the full activity of communication in five levels between a familiar person and the child.(48) We referred to children as having communication impairment if CFCS was level 4-5. Speech impairmentwere defined as inability to create or form speech sounds.(175)

Eligibility Criteria

Inclusion: Children aged 4 to 15 years diagnosed with CP (by a paediatric neurologist who would using standardized diagnostic criteria) of any type or severity. CVI and PVD were notcriteria for inclusion before randomization into the study (rational in introduction).

Exclusion: Children beyond the age criteria, with other causes of motor disorders, childrenwhose carers refuse to participate and children with CP who have debilitating illness and require immediate medical care.

Interventions Intervention arm

The intervention was the application of carer selected tailored IQI VSS(at maximum of 8) based on the “sometimes”, “often” or “always” response to the 52 IQI questions which they consider to be the most important, relevant and practical to implement. So if any one of the 52 questions in Insight was responded to as being a problem always, often or sometimes, a group of strategies to help adapt to this particular problem was suggested and explained to thecarer. If 6 questions were responded to as problematic, 6 groups of strategies would be administered to the carer. If more than 8 were responded to, the most relevant 8 problems/strategy groups chosen by the parents were selected for the carer to concentrate on. The strategies were explained at baseline and reinforced with phone calls.

The standard/control treatment arm

The types of challenges highlighted by the IQI are not assessed or treated routinely in management of CP in our environment, In the control arm, no vision support strategies weregiven after the IQI had been administered. After the 6 week follow up assessment had been completed, children in the control arm were offered IQI vision support strategies based on their response to the IQI.

Outcome

The primary outcome was change in quality of life, between baseline and follow up, assessedusing the PedsQL 4.0 Generic and PedsQL 3.0 CP modules. These outcomes were all compared between the intervention and standard treatment arms at six weeks.

The secondary outcome measure for visual function was the IQI mean scoreschange/difference from baseline to follow up.

Data collection methods

Data forms for socio-demography of carers and subjects were filled and analysed. The PedsQL 4.0 Generic, PedsQL 3.0 CP, IQI and IQI VSS, and follow up forms were used to collect data. Data were collected at baseline and at the end of follow up after 6 weeks. Data was entered into one Microsoft excel database by a masked medical records officer who wasnot part of data collection.

Quality of Life– Methodology

The quality of life methodology is described according to the format of the designers. For the PedsQL 4.0 Generic and the PedsQL 3.0 CP modules, the parent's proxy form was used. To create Scale Scores, the mean was computed as the sum score of the items over the number ofitems answered (this accounts for missing data). If more than 50% of the items in the scale are missing, the Scale Score should not be computed. Imputing the mean of the completed items in a scale when 50% or more are completed was the method used.

The PedQL 4.0 Generic module has 4 subscales which identifies problems with: 1) Physical functioning, 2) Emotional Functioning, 3) Social functioning and 4) School functioning.

The PedQL 3.0 CP module has 7 subscales which identifies problems in activities in everyday living, they are:1) daily activities 2) School activities, 3) Movement and Balance, 4)Pain and Hurt, 5) Fatigue, 6) Eating activity, 7) Speech and communication.

The school scores were not used as over 50% of children were not in school. To createthe Total Scale Score, the mean is computed as the sum score of all the items over the number of items answered on all the Scales.

The higher the score report the better the quality of life.

Sample size

Unpublished data from a pilot study in Bangladesh was used in the sample size calculation.In this study of 180 children with CP, the visual support strategies showed an impact on quality of life, measured by PedsQL 4.0 Generic of approximately 0.3 standard deviation (SD). (91)Using the Altman nomogram, a sample size of approximately 370 children with CP, with 185 children in each arm, is needed to detect an effect size of 0.3SD with 80% power and at a 95% confidence, allowing for 5% loss to follow up.

Randomization: sequence generation

The database of children recruited with information on their unique ID, age, sex and GMFCSwas sent to the data analyst in the University of Calabar Teaching Community Medicine Department at the end of the examination per local government area. The randomisation sequence was generated by the data analyst (SA) using Stata 11 programming syntax for block randomisation of patients into the treatment and control group. Children were stratified

and blocked by age groups 4-9 and 10-15 and by Gross Motor Function Classification Score (GMFCS) Levels 1-3 and 4-5.

Randomization: allocation concealment

Carers of children where contacted by an independent research clerk who was masked to baseline examination. Carers were invited to visit the same primary health centre where the baseline examination was conducted. At this visit, carers assigned to the treatment arm of thetrial received the intervention which was explained to them by the social workers while thosein that standard arm received counselling on observation of the child's symptoms and a final follow up call.

Implementation/Fidelity

Carers were to identify a maximum of 8 best strategies for implementation and were to conduct the

strategies three times daily. Carers were encouraged to start with the selection of1-3 of the most important strategies to them, to start with and practice the intervention three times a day, and those which could be implemented thereafter were identified and explained with each follow up phone call. Carers were given a list of all the strategies they had selectedto study further. To improve adherence and as part of the intervention, carers were contacted by phone call weekly for 6 weeks, to ask about the application of the strategy, the frequency of the application and if there were any side effects. Carers of children in intervention and control arms were unlikely to meet after the baseline assessment and allocation to arms, as each family was discharged individually and no physical group or internet based group was formed.

Blinding(masking)

A different set of social workers conducted the post intervention interviews. Allocation was concealed to the PI and all the members of the examination team who were different from those in the intervention and follow up team.

Follow-up

The trial period was 6 weeks, during which phone calls were made to monitor the progress of implementation of the visual support strategies, to identify additional new strategies for implementation for the following weeks, and to remind parents to visit the primary health centre for the final follow up. Follow-up interim calls were performed weekly for 6 weeks in the intervention arm. The standard treatment arm received a single call at the 6^{th} week as a reminder for the final follow up visit. Final interviews and data collection were conducted in the primary health centres for those that presented and in homes for those that could not present.

Informed Consent

The study was performed in accordance with the Helsinki declaration and approved by the ethics

committees of Cross River State and the London School of Hygiene & Tropical Medicine. A written information sheet was read out and informed consent was obtained fromall subjects under 18, from a parent and/or legal guardian.

Statistical methods

Statistical analyses were conducted with STATA (version 15.1) Analysis of data for parametric or non-parametric distribution was initially done. The T-tests was used to detect

difference in parametric data with a significance threshold at p=0.05. These were used tocompare intervention versus control group and also to assess the relationship between baseline characteristics and the primary outcome measure.

We analysed by intention to treat (the treatment group was considered as all those randomized to get IQI VSS), per IQI protocol (treatment group considered as those that actually required and received at least 1 VSS) and per VSS treatment (those that actually implemented at least 1 tailored VSS based on parent report) approach (as reported in the phone calls/at follow up), for all primary outcome analysis. Descriptive statistics includedmeans and SDs, and medians and interquartile ranges as appropriate. The number and proportions for categorical variables describing the sociodemographic details of the population by randomization were derived.

The impact of our intervention was measured using the difference in pre and postintervention scores.

The analysis of the IQI that showed responses with >/=50% as non-applicable and questions 9,5,15,32,33,34,45,46 were expunged.

Ethical approval

The study was performed in accordance with the Helsinki declaration and approved by the ethics committees of Cross River State and the London School of Hygiene & Tropical Medicine. A written

information sheet was read out. Informed consent was obtained from

parents and /legal guardian. Children were referred for health care services as needed.

Results

Participants flow

A total of 1024 children were identified by key informants, 293(28.6%) were not brought for examination. 731 children were assessed for cerebral palsy diagnosis eligibility, 343(46.9%) did not meet the inclusion criteria for CP and 388(37.9%) were confirmed to have CP; all enrolled into the RCT, no parent declined to participate. 388 children who met the criteria forCP were then randomized; 191(49%) were allocated to intervention while 197(51%) were allocated to standard intervention. Of the 191 that were allocated to intervention, 44(23%) ofchildren did not meet the criteria for requiring VSS treatment as they did not answer, ‘always’, ‘often’ or ‘sometimes’ to any of the Insight Questions Inventory questions.

Follow up phone calls in the intervention arm was made in the following frequency: week 1: 99(26%), week 2: 96(25%), week 3: 82(21%), week 4: 71(18.3%), week 5: 55(14%) and week 6: 37(10%). However, the key informant method ensured that the key informants kneweach child’s home and was able to ensure they attended the follow up.

383 (98%) children completed the study protocol, and analyses performed for the primaryand secondary outcomes. Figure 1 shows the consort flow chart.

Recruitment

Children were recruited per local government area into the study from December 2016, andallocated to an intervention arm or a standard treatment arm. (Table 1) The study data collection was closed in December 2018.

Baseline data

Baseline demographic and clinical characteristics of each group can be seen in Table 1. Table 2 shows the distribution of the place of residence and father and mothers' educationalstatus of children with CP. 89% of children were from rural areas, 30% of mothers and 29%of fathers were subsistent farmers.

A total of 331(85.3%) children had speech impairment and 173(44.6%) had communication impairment.

A total of 335/388(86.3%) children had at least one symptom of PVD in this population, 147/191(77.0%) in the intervention arm which required the administration of at least one Insight Questions visual support strategy and 188/197(95%) in the control arm, p= 0.0012.

We assessed 6 factors at baseline for their effect on QoL; residence (urban vs rural), sex (male vs female), age (<9years vs >9 years), type of CP (spastic vs others), GMFCS (ambulatory I-III- vs non-ambulatory IV-V), and presence or absence of PVD (according to the IQI). Baseline QoL comparisons showed that those with non spastic CP had a better QoL(mean 43.1 vs 39.2, p= 0.0337 for PedsQL 4.0 Generic and mean 58.2 vs 48.8, p= 0.001 for
3.0 PedsQL CP) ; also for PedsQL 3.0 CP only, children older than 9 years had a higher meanand better QoL (56.9 vs 45.4 p< 0.001).

There was a baseline difference in QoLbetween the two arms with the PedsQL 4.0 Generic,with children in the treatment arm having better QoL (42.1 vs 38.7, p=0.0411).

Numbers analysed

Three hundred and eighty-eight children were randomized and 191/388 (49%) allocated to intervention and 197/388(51%) allocated to standard treatment representing the number in the

intention to treat analysis.

For the per IQI protocol analysis, 147/191(77%) met the criteria for the IQI protocol administration of intervention, as forty-four children in the intervention group did not havesymptoms of PVD in any subdomain and therefore did not get strategies. For the per VSStreatment analysis, there were 91/191(48%) participants randomized to treatment who actually implemented the strategies three times a day.

The mean number of strategies assigned to each family was 3.7(SD 2.9)

The first three choices of strategies with the highest frequencies chosen by parents were to Insight Questions and strategy numbers 17:100/147(68%); If the child has difficulty catchinga ball: ❶Practice catching skills with your child by throwing a balloon to each other. The balloonwill move slower than a ball and may be easier for your child to catch. ❷Put a little bit of rice / water in the balloon. The balloon will make a noise as it moves so your child can hear where it is. ❸Use large, brightly coloured balls when playing catch or other ball games with your child. ❹Use balls with sound or light effects when playing catch or other ball games with your child.

Number 4:52/147(35%); If the child appears to 'get stuck' at the top of a slide or hill:

❶Encourage your child to practice around the house trying to cross small gutters and playing on small play slides and/or by lying on his tummy on a scooter board or skate board.Some children choose to go down slides head first. Do not stop this, but make sure it is safe.
Children may do this because the upper part of their field of view is being used in thissituation.
❷Give additional verbal information.

Number 37:40/147(27%).

If the child reacts angrily when other restless children cause distraction:

❶If possible, take other distractions away from your child's work area. i.e. sound, movement.

❷Let your child use head phones or ear plugs so noise does not disturb him.

❸See how your child gets on sitting at a separate desk at the end of the group. This maygive him more space without leaving him out of the group.

Outcomes and estimation

For the primary outcome analysis, the results of 'intention-to-treat', 'per IQI protocol' and 'per VSS treatment' analysis did not show any significant difference between intervention and control groups, in change or improvement in the total quality of life in children using either the generic or the PedsQL-CP tool using the unpaired t test. (Table 3 and 4 shows the intention to treat analysis, per IQI protocol and per VSS implementation for the Total CP generic and CP scores). Testing indicated that all the data were parametric so t-test were usedthroughout.

Table 5 shows the subdomains of speech and communication, where there was a betterimprovement in quality of life in the intervention group compared to the control group,analysed by intention to treat and per IQI protocol. ($p=0.035$ and $p=0.006$)

The secondary outcome measure (the Insight Questions Inventory) is reported in Table 6 as intention-to-treat, per IQI protocol or per VSS implementation. There was no overall difference between intervention and control groups.

Adverse events

Two patients died in the standard treatment arm and one in the intervention arms of the studydue to complications of CP. There were no side effects reported from any group.

Discussion

In this community based RCT, visual support strategies aimed at compensating for visual perceptual problems identified by the Insight Questions inventory (IQI VSS) were not shownto significantly improve the overall quality of life of children. The IQI tool did not function effectively for this population. It did not elicit sufficient positive responses in children who seemed to have evidence of PVD using some basic objective tests.(95) Since the IQI VSS depends on such positive responses this would have limited its effectiveness. IQI did seem tobe effective, in a similar population in Bangladesh, in eliciting symptoms which did relate to quality of life; and IQI VSS have shown some promise in improving quality of life in a UK hospital based study of children with CVI.(92) Although it has been successful in other studies it should be noted that the IQI questions taken individually are not specific for CVI orPVD: they could be answered positively by children with ocular VI or pure motor impairment.

Further work is required refining this tool for this population including investigation of which questions are appropriate, for children with CP/comorbidities, for families in more rural environments, and for parents with a range of education levels. It is possible that a questionnaire approach is not the best one for this population. Separate from how the IQI

performed we did find evidence of under recognition / reporting of visual morbidity by the carers of these children (95). In retrospect it could be argued that we should have screened forCVI or PVD before recruiting e.g. perhaps had a cut off requiring a certain number of positive IQI responses to be eligible for recruitment. Our assumption was that almost all children with CP have some level of PVD but this proved not to be the case; almost a quarter did not receive any strategies which resulted in an underpowered trial. The results might havebeen influenced by chance differences between the 2 groups at baseline. There was a higher proportion of PVD in the control group compared to the intervention group. Also the treatment group had better scores PedsQL 4.0 scores at

baseline though this was a small difference and would lose significance after adjustment for multiple comparison (p=0.04 and we tested differences in PedsQL 4.0 and 3.0 so threshold would reduce to p=0.003).

Another reason for the lack of effect could have been poor adherence to the strategies. Therate of successful follow-up calls dropped substantially across the 6-weeks intervention period, it is possible that parent implementation of strategies decreased and that adherencemight have been over reported.

Improvement due to the use of IQI visual support strategies, in the health related quality of life in the CP speech and communication domain was suggested across the intention to treat and per IQI protocol. However, this may be a chance finding as our analysis was not adjustedfor multiple comparisons. (Since there were 2 PedsQL modules each with subsections, a threshold value p value of 0.05, for instance, would drop to 0.003) The impact of the VSS on speech and communication needs further investigation before drawing strong conclusions andshould be further investigated since a large proportion of children in this population had speech impairment (85%) and 45% had communication difficulties. It is well reported that

these are common problems in children affected by CP (211). Various strategies have beentried, but evidence of their effectiveness is limited.(211)

A surprising finding was that quality of life seemed to improve more in the control arm than the intervention arm when the PedsQL 4.0 Generic score was used whereas the reverse was true when the PedsQL CP 3.0 module was used. The differences were not significant and may be due to chance but this difference in trend is notable and may be because of the differences in the 2 tools with the CP module being more physical disease focused and easierfor parents to relate with their child's problems. Another possible reason for the discrepancy between the 2 PedsQL versions is

that the main benefit was seen in the speech and communication domain of the PedsQL 3.0 CP tool which is not present in the generic version. Some children appeared to have a decrease in quality of life in the 4.0 Generic module following the intervention. It is possible that having undergone lengthy assessments and not received any medical treatment or glasses immediately, carers may have felt a lack ofintervention or improvement and reflected this in their answers. In addition, they may have thought more (after the first experience of answering the PedsQL questionnaires) about problems children have, so give more negative results second time around, a possible situation of investigation fatigue (this may have also contributed to the lack of improvement in IQI scores in the treatment group, where previous work did find such an improvement)(92).

Limitations of the study

A rigorous RCT was conducted in a field where there is little RCT evidence on which to base management of this condition. However, the assessment and intervention tool did not seem towork well in this population, despite having worked well in a range of geographic

populations previously. IQI scores and the number of children receiving IQI VSS were smaller than predicted, likely underpowering the trial. More cultural adaptation and pilotingwould have been beneficial such as checking whether the questions were appropriate (for children with CP/comorbidities, for families in more rural environments and for parents withthe education levels noted).

Conclusion

The use of the IQI and IQI VSS for the treatment of PVD in children with CP in this population did not show any superiority over current standard measures of treatment. The study suggests that further investigation and refinement of this type of intervention is required for this population.

There was a suggestion of a positive effect in the area of speechand communication related quality of life.

Table 9.1: Sociodemographic and clinical characteristics of the intervention and nointervention arms (N=388)

Characteristics		Intervention arm		Control arm	
		No	%	No	%
Total		191	100.00	197	100
Local government area					
	Abi	3	1.6	2	1.0
	Akamkpa	6	3.1	8	4.1
	Akpabuyo	8	4.2	13	6.6
	Bakassi	6	3.1	5	2.5
	Bekwara	15	7.8	13	6.6
	Biase	8	4.2	11	5.6
	Boki	15	7.8	16	8.1
	Calabar Municipality	7	3.7	3	1.5
	Calabar South	13	6.5	19	9.6
	Ikom	10	5.2	10	5.2
	Etung	7	3.7	8	4.1
	Obanliku	13	6.8	14	7.0

	Obubra	9	4.7	11	5.6
	Obudu	15	7.8	12	6.0
	Odukpani	3	1.8	4	2.0
	Ogoja	25	13.5	19	9.6
	Ugep	8	4.2	8	4.1
	Yala	20	10.5	21	10.6
Residence					
	Urban	21	11.0	23	11.7
	Rural	170	89.0	174	88.3
Sex					
	Male	120	62.8	109	55.3
	Female	71	37.2	88	44.7
Child's age					
	Mean (SD)	9.08	4.0	9.2	4.0
	Median (IQR)				
	<9 years	98	51.3	114	57.9
	9+ years	93	48.7	83	42.1
GMFCS					
	1	35	18.3	35	17.8
	2	80	41.9	76	38.6
	3	22	11.5	32	16.2
	4	30	15.7	24	12.2
	5	24	12.6	30	15.2
GMFCS					

	Ambulatory	137	71.7	143	72.6
	Non ambulatory	54	28.3	54	27.4
Anatomic					
	Monoplegia	18	9.4	13	6.6
	Triplegia	26	13.6	21	10.7
	Diplegia	10	5.2	9	4.6
	Hemiplegia	70	36.6	80	40.6
	Tetraplegia	67	35.1	74	37.6
CP Type					
	Spastic	137	71.7	134	68.0
	Ataxic	20	10.5	18	9.1
	Dystonic	10	5.2	8	4.1
	Unclassified	13	6.8	19	9.6
	Choreoathetoid	11	5.8	18	9.1
Visual acuity (Mirror Test)					
	Normal	92	48.2	83	42.1
	Visual impairment	99	51.8	114	57.8
PVD(IQI)					
	PVD	147	77	188	95.4
	No PVD	44	23	9	4.6
PVD(tests)					
	PVD	86	48.6	91	51.4
	No PVD	105	51.4		48.6

Table 9.2: Distribution of the place of residence and father and mothers' educational status of children with cerebral palsy (N=388)

Variables	No	Percent
Fathers age (n=353)	Mean age 42.7(SD 15.5); Median 40;(IQR35,50)	
Mothers age (n=373)	Mean age 34.3(SD 8.4); Median 32 (IQR 29, 40)	
Place of residence		
Urban	44	11.3
Rural	344	88.7
Educational status		
Fathers educational status		
No formal education	19	4.9
Incomplete primary education	7	1.8
Completed primary	56	14.4
Completed junior secondary	15	3.9
Completed senior secondary	129	33.2
Post-secondary education	79	20.4
Post graduate education	2	0.5
Could not be ascertained	81	20.9
Total	388	100
Mothers educational status		
No formal education	17	4.4
Incomplete primary education	16	4.1
Completed primary	81	20.9
Completed junior secondary	26	6.7

Completed senior secondary	150	38.7
Post-secondary education	60	15.5
Post graduate education	1	0.3
Could not be ascertained	37	9.5
Total	388	100
Occupational/Skills		
Mother		
Unskilled (subsistence farmers)	118	30.4
Semiskilled	233	60.0
Civil/Public servant	28	7.2
Professional	9	2.3
Father		
Unskilled (subsistence farmers)	112	28.9
Semiskilled	196	50.5
Civil/Public servant	66	17.0
Professional	14	3.6
Income/month		
Fathers income n=353	Mean $13.4; Median 0; IQR(0,0)	
Mothers income n=373	Mean $13.1; Median 0; IQR(0,5.3)	

Table 9.3: Primary outcome measure, total PedsQL 4.0 Generic scores using intent-to-treat (N=388), per protocol (N=344), per treatment analysis (N=288)

Variable	Intention to treat			Per IQI protocol			Per VSS treatment		
	ntervention arm	ontrolarm	P value (t test)	ntervention arm	Control arm	P value (t test)	ntervention arm	Control arm	P value (t test)
Total PedsQL 4.0 baseline								0.3234	
N	191	197		147	197		91	197	
Mean (SD)	42.1(SD 19.5)	38.7(SD 18.6)		43.2(SD 18.3)	.7(SD 18.7)		41.2(SD 17.5)	38.7(SD 18.6)	
Median (IQR)	41.7(25,56.25)	37.5(25,54.2)		43.7(27.1,56.2)	37.5(54.2)		41.7(27.1,52.1)	37.5(25,54.2)	
Total PedsQL									

4.0 follow up									
N	157	157		125	157		91	197	
Mean (SD)	45.1(SD 22.9)	44.1(SD 21.6)		45.5(SD 21.9)	4.1(SD 21.6)		41.2(SD 17.5)	38.7(SD 18.6)	
Median (IQR)	43.7(27.1 ,58.3)	41.7(29.2 ,58.3)		43.7(29.5 ,58.3)	41.6(2 9.2, 58.3)		41.7(27.1 ,52.1)	37.5(25, 54.2)	
Total PedsQL 4.0 differ ence			0.9 43			0.9 317			0.1 00
N	157	157		125	157		81	157	
Mean (SD)	2.3(SD 11.8)	5.0(SD 17.8)		2.3(SD 11.8)	5.0(S D 17.8)		2.3(SD 10.0)	5.0(SD 17.8)	
Medi an (IQR)	9(0,0)	0(0,0)		0(0,0)	0(0,0)		0(0,0)	0(0,0)	

Table 9.4: Showing the results for the primary outcome measure on the total PedsQL 3.0 CPscores using intent-to-treat (N=382), per protocol (N=339) and per treatment analysis (N=283).

Variable	Intention to treat			Per IQI protocol			Per VSS treatment		
	ntervention arm	ontrolarm	P value(t test t)	ntervention arm	Control arm	P value (t test t)	ntervention arm	Control arm	P value (t test t)
Total PedsQL 3.0 baseline									
N	189	193		146	193		90	193	
Mean (SD)	53.5(SD 26.5)	49.8(SD 27.6)		53.9(SD 26.3)	49.8(SD 27.6)		52.4(SD 26.7)	49.8(SD 27.6)	
Median (IQR)	55.5(34.7,76.1)	45.2(27.4,74.2)		53(34.7, 76.6)	45.2(27.4,74.2)		47.6(33.9,74.2)	45.2(27.4,74.2)	
Total PedsQL									

3.0 at follow up									
N	164	162		130	162		86	162	
Mean (SD)	60.4(SD 27.2)	54.4(SD 27.5)		62.3(SD 26.9)	54.4(SD 27.5)		61.6(SD 27.8)	54.4(SD 27.5)	
Median (IQR)	61.7(42.2,83)	53,2(32.2,77.4)		63.3(43.5,87.1)	53.2(32.2,77.4)		64.9(41.9,87.1)	53.2(32.2,77.4)	
Total PedsQL 3.0 difference			0.287			0.163			0.908
N	163	161		130	161		86	161	
Mean (SD)	6.3(SD 22.2)	4.9(SD 23.2)		7.5(SD 22.4)	4.9(SD 23.2)		9(SD 9)	4.9(SD 23.2)	
Median (IQR)	0(-2.4,19.5)	0(0,13.7)		0(-1.6,20.1)	0(0,13.7)		0(-2.4,27.4)	0(0,13.7)	

Table 9.5: Primary outcome measure, paediatric quality of life cerebral Palsy speech and communication subdomain scores for using intent-to-treat (N=310), per protocol (N=247),per treatment analysis (N=227)

Variable	Intention to treat			Per IQI protocol			Per VSS treatment		
	Intervention arm	Control arm	P value (t test)	Intervention arm	ontrol arm	P value (t test)	Intervention arm	ontrol arm	P value (t test)
PedsQL3.0 Speech and communication at baseline									
N	154	156		118	156		71	156	
Mean (SD)	44.1(SD 40)	42.9(SD 41)		42.3(SD 40)	42.9(SD 41)		43.3(SD 41.4)	42.9(SD 41)	
Median (IQR)	37.5(0,81.2)	31.2(0, 94)		34.3(0,81.2)	31.2(0, 94)		37.5(0,93.7)	31.2(0, 94)	
PedsQL3.0 Speech									

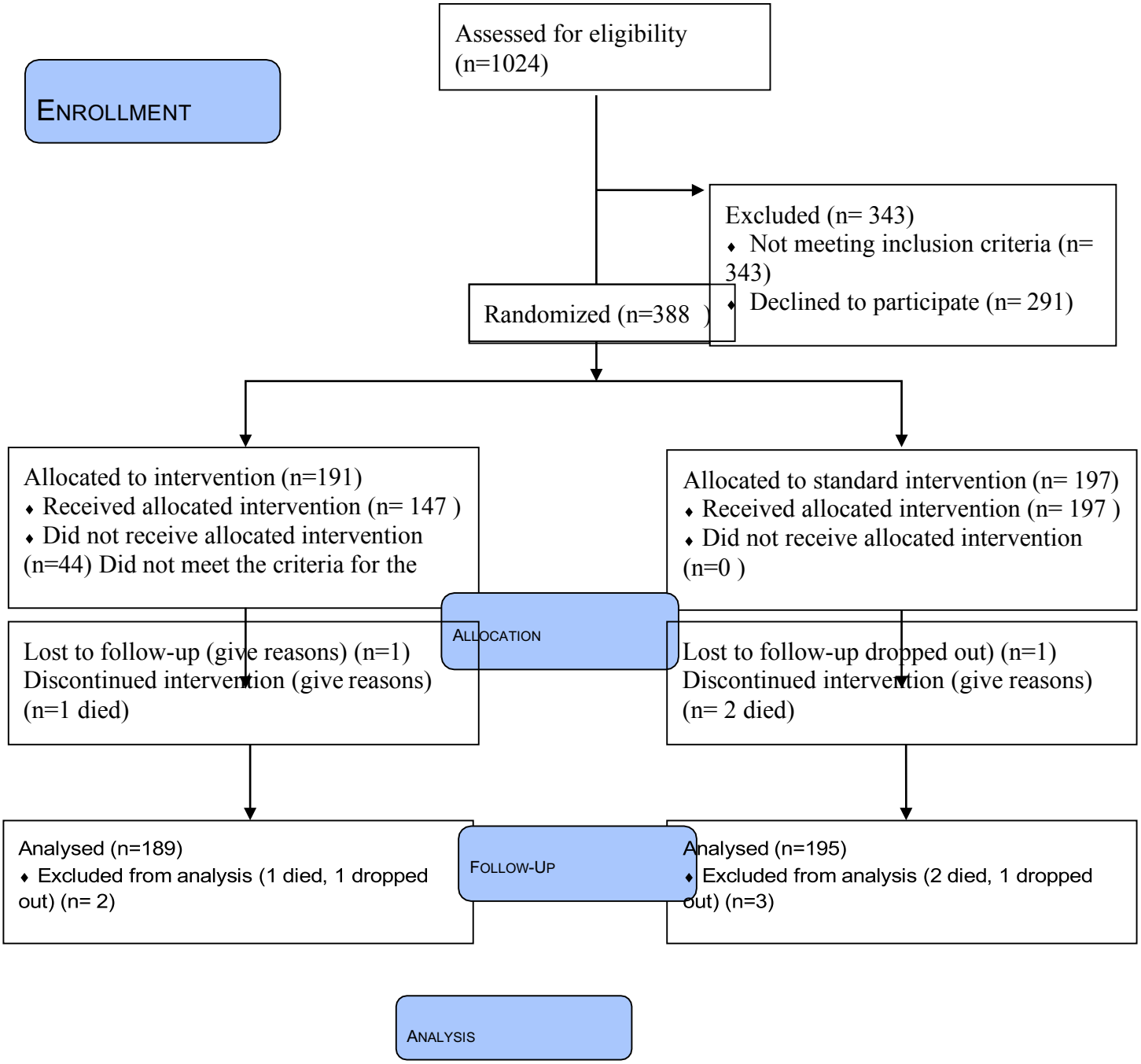
and communication at follow-up									
N	139	136		110	136		71	136	
Mean (SD)	51(SD 43.1)	42.2(SD 41)		53.6(SD 43.6)	42.2(SD 41)		55(SD 44.4)	42.2(SD 41)	
Median (IQR)	50(0,100)	25(0,93.7)		50(0,100)	25(0,93.7)		62.5(0,100)	25(0,93.7)	
PedsQL3.0 Speech and communication difference									
N	133	130	0.0350	105	130	0.006	66	130	0.994
Mean(SD)	7(SD 35.2)	-0.64(SD 32)		10.5(35.7)	-0.64(SD 32)		12.4(SD 46.8)	-0.64(SD 32)	
Median (IQR)	0(0,18.75)	0(0,0)		0(0,025)	0(0,0)		0(0,31.2)	0(0,0)	

Table 9.6: Secondary outcome measure of visual function using the Insight QuestionsInventory using intent-to-treat (N=383), per protocol (N=341), per treatment analysis(N=285)

Variable	Per treatment			Per IQI protocol			Per VSS treatment		
	tervention arm	Control arm	P value(t test)	Intervention arm	Control arm	P value (t test)	Intervention arm	Control arm	P value (t test)
Total IQI score at baseline									
N	188	195		146	195		90	195	
Mean (SD)	1.5 SD(0.5)	1.7 SD(0.7)		1.6 (SD 0.48)	1.7 (SD74)		1.64(SD 0.54)	1.67 (SD 0.74)	
Median (IQR)	1.3(1.2, 1.7)	1.4(1.2, 1.9)		1.4 (1.2,1.7)	1.4(1.2 ,1.9)		1.5(1.3 ,1.8)	1.4(1.2, 2.0)	
Total IQI follow up scores									
N	188	192		146	192		90	192	

Mean (SD)	1.2 SD(0.62)	1.3 SD(0.8)		1.25 (SD0.57)	1.3 (SD0.8)		1.36(SD 0.51)	1.34 (SD 0.81)	
Median (IQR)	1.3(1.0, 1.5)	1.2(1.0, 1.1.6)		1.2(1.0, 1.5)	1.2(1.0 ,1.66)		1.3(1.0 , 1.56)	1.3(1,1, 1.67)	
Total IQI scores of difference			0.322			0.65 9			0.56 7
N	188	192		146	192		90	192	
Mean (SD)	-0.27 SD (0.61)	-0.3 SD (0.8)		-0.33(S D 0.6)	-0.30(S D0.8)		-0.29(S D 0.51)	-0.30 (SD 0.79)	
Median (IQR)	-0.13(-0.4,0)	-0.12(-0.4,0)		-0.15(-0.3, -0.5)	0(-0.1,0)		-0.17(-0.4 - 0.05)	0(-0.17 , 0)	

Figure 9.1: CONSORT flow diagram for the effectiveness of visual support strategies for visual impairment in children with cerebral palsy showing the enrolment, intervention andassessment.



10 Discussion (Thesis)

Cerebral Palsy being the most common motor disability in childhood, including in Africa andNigeria, requires extensive studies, especially in lower middle income areas where there is a paucity of information.(192, 212) Furthermore, the associated conditions such as abnormalities in perception , including visual perception, has become the leading cause of childhood visual impairment in high income countries and lower middle income countries with very little known about the condition..(28, 55, 139, 213) This thesis aimed to gain detailed clinical descriptions of cerebral palsy in our population, with regard to aetiology, ambulation, co-morbidity, visual impairment and access to education. We also aimed to assess the effectiveness of the IQI strategy (structured clinical history inventory with associated tailored visual support strategies) in improving quality of life in children with CP in our population, through a randomized control trial. In order to do this, we needed to make an attempt at appropriate cultural and linguistic modifications to the tool, the Insight questions inventory.

This chapter on discussion highlights the main research findings from each research chapter and describes the strengths, weaknesses, and conclusions of this work. The implications of these findings and future research directions in cerebral visual impairment in children with cerebral palsy.

In summary, the trial protocol was registered in the Pan African Clinical Trials Registry(PACTR) with number PACTR201612001886396 (chapter 4). Attempts at cultural/adaptation/translation showed that parents generally under recognized and underreported symptoms of perceptual visual dysfunction, however, the Nigerian Version of the IQI with the complete 52 set of questions retained, was produced (chapter 5). We found aprevalence estimate of 2.3 (95% CI 2.0-2.5) per 1000 children within this region of Nigeria (Chapter 6) Furthermore, the three commonest associated comorbidities in this population

were, learning difficulties 88%, feeding difficulties 86% and speech impairment 85%. Poor school attendance was noted. The study showed that binocular visual acuity impairment (BVAI) was seen in 20/201 by Lea symbols test (10%), and 213/388 (55%) by the mirror test, and yet visual problems were reported by carers in only 14%. The estimated frequency of cerebral visual impairment (CVI) in children ranged from 61(16%), to 191(49%) if children with optic atrophy were included. The trial showed The use of IQI VSS for the treatment of PVD in children with CP in this population does not show any superiority over current standard CP management in terms of overall quality of life. However, there was some evidence of improvement in quality of life in the area of speech and communication.

10.1 Discussion on the main research findings

10.1.1 Translation and cultural adaptation of the Insight Questions Inventory and visual support strategies.

The IQI/VSS was developed in Britain for an English population, and had not been used in Africa, including Nigeria. To overcome misunderstanding of inventory, it was necessary to adapt the inventory to the Cross River State, Nigeria population to align with cultural differences and language. Results show that the IQI can be adapted to a different population. Specific changes were made to the tool in cultural descriptions of the environment such as a hill versus a staircase and also took into account language expressions such as to ‘spot’ ratherthan to ‘find’. A few questions relating to the use of technology were seen not to be very useful in this population children, however since the inventory was for the general population, it is assumed that it should be able to cater for everyone, hence the questions were left in the modified Nigerian inventory.

Also noted was the under recognition and reporting of signs of PVD using IQI in this population. Educational programmes towards training in the identification of PVD symptoms is needed for this population. The main limitation of this pilot was the participation of a small number of carers who were mainly from an urban area in the process.Hence a true representation of the rural setting was not reflected, as seen in the study were 88.6% of the children with CP were from rural areas. In addition, the visual support strategiesdocument was given in English even though explained in verbally in the local language. A document with visual aids may have been more useful to enable better understanding and recall of strategies in the parents. The British IQI version and visual support strategies is generally used in high income countries, however, a high proportion of parents (father 80% and mother 85% have less than post-secondary education) were in the lower socioeconomic category in our population. This may also have contributed to the challenge of under recognition and under reporting that was seen.

Further work on whether a structured clinical question inventory such as IQI would be useful in managing this condition in this population, and if so, considerable work would be needed to make it more effective.

10.1.2 Clinical features and aetiology of cerebral palsy in children from Cross River State, Nigeria

Similar to other studies in developing countries, the use of the KIM to identify children with CP in Nigeria is feasible and yields a good result. The use of this method in a lower middle income setting also has implications for research, service provision and delivery and the development of a cerebral palsy register in this country. Data on prevalence and type of CP insub Saharan Africa are lacking. Our study yielded a prevalence estimate of 2.3 (95% CI

2.0-

2.5) per 1000 children within this region of Nigeria (Chapter 6), compared with a recent population based survey of (2·8 (2·4–3·6) per 1000 children, from Uganda.(29) We understand the stigma associated with CP and children with disability in general in our population and our study suggested that several children who were suspected to have CP were not brought to the health centers for assessment (Key informants identified 1024 children, 293(28.6%) were not brought for examination, 344(33.6%) did not have CP and 388(37.9%) were confirmed to have CP. The results also showed a similar profile of greater severity as most studies in Africa has shown. However, it is possible that the non -attendance mentioned above may have biased our sample in terms of gender, severity, type of CP and co-morbidities and population based studies continue to be useful in addition to KIM methodology for this reason. Preventable causes of CP in these Nigerian children were identified including hyperbilirubinemia and congenital rubella syndrome. Congenital Rubellasyndrome only requires a vaccine programme to eliminate CP from this cause. This vaccine programme is still absent in Nigeria. An important limitation of this retrospective analysis was the reliance on parent reports for attributing aetiology. Despite this, the result is similar to that of other countries and contributes to knowledge and the need to focus and advocate onthe prevention of causes of neonatal morbidity in this population especially from avoidable causes such as hyperbilirubinemia for which health promotion and education programmes as well as management strategies have been established for. (214)

10.1.3 Pattern of comorbidities in school-aged children with cerebral palsy in CrossRiver State, Nigeria

The definition of CP clearly includes the consideration of the associated comorbidities that

exist in this children as part of the CP disorder. Our research was able to identify key comorbidities in this population, showing that CP in this population is a multimorbid condition with untreated comorbidities. Much clinical effort has been put into ‘treatment’ of impairments, and assumed that improvements in body structure and function would make a child better and thus would lead to functional gains. Following the new way of thinking along the line of the ICF classification, more attention should be given tothe functional implications of the co-morbidities in addition to medical management where appropriate.

Learning disability was the commonest associated impairment and this was seen in a similar study in Uganda. (30) Speech and epilepsy were also very common impairments with epilepsy the main reason for hospital attendance. (32)

During the analysing of the PedQL4.0 and 3.0 quality of life tools, the school analysis had to be expunged because over 50% of children were not participating in any form of education orschooling. Comorbidities significantly associated with lack of schooling were visual impairment, epilepsy and learning disability. Interventions may help for some of these conditions. However, severity of CP measured by ambulation was the second most significant barrier to school attendance, emphasising the need to facilitate school attendance by addressing the need for ambulation in children with CP or mobility of children to school through transportation support. Education is one of the rights of a child, hence lack of school attendance infringes the rights of children with CP and needs to be focused on in this population. The myriad of comorbidities suggests an unmet health services need; an example of this is epilepsy which was untreated in about 95% of children who had the condition in ourpopulation.

Nigeria is one of the 3 high burden countries where the Survive & Thrive Alliance works

with country governments and health professionals in countries throughout Asia and Africa to improve health outcomes for mothers, children, and new-borns through clinical training, systems strengthening, and policy advocacy. It is expected that with this support that some aspects of the eating of CP which involves maternal and neonatal and infant mortality will beaddressed. Major limitations of this study included the inability to use standard methods of clinical assessment for some of the associated conditions such as learning disability and also the inability to investigate the children with technological advanced tests such as the magnetic resonance imaging.

As an ophthalmologist, I took one of the co-morbidities, visual impairment and assessed this in more detail.

10.1.4 Visual impairment in children with cerebral palsy in Nigeria

High levels of visual morbidity were found, generally under-recognized by carers.

The study illustrated that visual assessment in children with CP requires a choice of the most suitable test that the child can cooperate with, not merely for the chronologic age but a test more suitable for their developmental age and capacity. Hence our report of the three types oftests conducted to assess and estimate the binocular visual acuities in this children. Even though children had visual impairment, yet visual problems were reported by carers in only 15% of cases. This under-recognition was not simply a failure of the IQI tool to pick up PVD, but was also demonstrated through responses to an open question about visual problems from carers of children with easily measurable visual impairment. With regards to the causes of the visual impairment, CVI was the commonest with aprevalence estimated at 49% (when children with optic atrophy, acommon epi-phenomenon in CVI were included) One type of CVI, PVD was investigated in detail despite lack of standardized tests,

particularly for this population. In this research, two methods were used to assess PVD; a subjective method using a behavioural tool the Insight Questions Inventory and an objective method by the use of tests (LEA visual assessment system). Scores on both these tests indicating presence of PVD both were seen to be associated with 1) abnormal behaviours (such as difficulties with, emotions, concentration, behaviour or getting on with other people or often loses temper) and 2) abnormal saccadic eye movements, indicating the close link between the sensory and motor aspects of vision in CVI. PVD was seen to occur both in children with normal visual acuity (as has been documented by other researchers),(124) and also seen in children with visual acuity impairment. Comparison of objective versus, subjective assessments for PVD suggested systematic underreporting by carers of problems relating to PVD in everyday life using the IQI, especially compared to similar populations in the UK and Bangladesh. This may limit its use as a tool for providing tailored visual support strategies for this population. The lack of awareness of vision problems by carers, was not, asalready mentioned, solely ascribable to failure of the IQI tool and highlights the need for health education about vision including PVD and for comprehensive visual assessment of these children. The data show a considerable burden of visual impairment within this group. We suggest that children with CP should undergo full ophthalmological assessment looking for impairments in visual acuity and field, contrast sensitivity and visual perceptual problems.This we recommend should not be conducted as a screening exercise because the impairments that are seen in this children are seen to be varied, graded and are not binary, and there are no well validated screening tools for CVI as yet. A comprehensive visual assessment of all children with CP is therefore mandatory.

10.1.5 Effect of visual support strategies on the quality of life of children with cerebral palsy and cerebral visual impairment/perceptual visual dysfunction in Nigeria: result of a randomized controlled trial.

Although benefit in QoL has been demonstrated in a small uncontrolled pilot study in the UK, (92) the IQI/VSS had not undergone the process of a randomized control trial. This research showed that the Insight Questions Inventory and visual support strategies compared to standard eye care treatment for the management of PVD was ineffective in improving the quality of life of children with CP in our population through an RCT and further work is necessary to find effective strategies for managing visual problems in these children.

10.2 Summary of findings

The overall picture is that CP is prevalent and severe with multiple co-morbidities which impair access to education additionally to motor difficulties. Visual impairment is a major morbidity affecting both acuity and higher order visual perception. There is systematic under-recognition of visual problems among carers. The majority of the visual impairment resulted directly from the brain injury as opposed to co-existent ocular morbidity. The RCT aimed at improving the quality of life and functional vision and based on parental reports of visual problems did not show improvement in the total scales of primary outcome (PedsQL 4.0 Generic and PedsQL 3.0 CP) and effectiveness in improving quality of life.

Further improvement in the use of the IQI to assess children with PVD might be beneficial As the body of research around PVD in general and in children with CP specifically continues to increase, further advances may be made. Further investigation is required of locally identifiable symptoms and behaviours of PVD for which locally conceived and appropriate questions and strategies may be suggested.

10.3 Diagnostic challenges

10.3.1

1) Limitations if IQI

Despite the gains of this investigation, further research is needed to fully understand PVD in this population and refine intervention strategies. The identification of few children with PVD using the IQI compared to the children identified with tests for PVD suggests poor utility for this method and the need for further development. The study also showed the challenge carers have in recognition, understanding, interpretation and reporting symptoms ofPVD. This maybe due to lack of knowledge as it as new are in medicine, or due to cultural bias of which carers are unwilling to add negative labels to child unless there is a clearly defined perceived benefit. There may also be difficulties distinguishing visual from motor or cognitive impairments – such difficulties may be experienced by health professionals and therefore inevitably also by parents and carers. This problem is not unique to LMICs but is seen in the UK as well. Some have suggested that formal neuropsychological tests are necessary to diagnose CVI of the PVD type but even there, access to the professionals neededto administer these tests, one of the reason why alternative clinical assessments such as IQI have been tested.

2) Lack of best corrected VA

Due to the difficulties in examining children with multiple disabilities in addition to the changing visual perception of children during the course of a day, and the distress most of thechildren faced with the use of the trial spectacles and the short time that was available to

undergo numerous examination, uniocular assessments and the best corrected visual acuity could not be performed, impairing a precise estimate of visual acuity impairment. However since very few of the children had access to refraction and glasses this does give an accurate picture of 'actual' vision in the community.

3) Problems measuring acuity

Just as our IQI responses were much more negative than Bangladesh, so our proportions of children able to perform preferential looking acuity were much lower. Fortunately, the mirrortest worked well in a discriminating way. This method could be further expanded by development of recognition visual acuity measures that make use of reflective ocular movements. Further development of such an assessment tool may eliminate a few limitations such as poor attention, communication and learning difficulties in the ascertainment of visualacuity in these children.

10.3.2 Scope of other interventions

The multimorbid nature of CP requires a holistic approach and a multidisciplinary approach to care. Ambulation was a major recurring association with PVD and neuro-ophthalmic abnormalities. The use of mobility aids is encouraged. . Environmental modification should be given priority in public spaces as well as homes to enable ambulation and mobility.

10.3.3 Implications of findings for helping children with CP in Nigeria

Reports are needed to sensitize governments of the urgent need to speed up the implementation of rubella vaccinations. Our report would contribute to the advocacy requiredto institute the rubella vaccination. Co-morbidities were shown to be associated with

poor school attendance and require medical and habilitational intervention. Our trial for improvingquality of life through vision support strategies did not show effectiveness and further research is required into how best to assess and mange this particular co-morbidity. There were high levels of speech, language and communication problems are there was some weak evidence that VSS may have an impact here, illustrating that vision may impact all areas of a child's development and perhaps this area in particular.

CVI clinics could be developed now with better understanding of this structured history inventory, commencing the process of identifying more local PVD presentations and strategies and parental education of CVI/PVD especially using photographic descriptions canbe done.

10.4 Implications of findings for future operational research priorities for children with CP inNigeria

The high prevalence of CP in this population suggests that the link between neonatal morbidity and CP may need to be further investigated. Clinical indicators for neonatal morbidity are available, such as the neonatal morbidity range, Morbidity Assessment Index for Newborns, and The Neonatal Adverse Outcome Indicator amongst others. However, a community or population indicator for the impact of neonatal morbidity is required, such as the use of the prevalence and incidence of CP. Ambulation is a major concern in children with CP, further research in the use of mobility aids and barriers to the use in children with CP needs to be investigated. Investigation of barriers to the development of intervention programmes for children with CP is urgently needed, to facilitate the institution of intervention programmes and learning for children in this population.

Development of an early intervention programmes for children and forming a parents' association to educate and train parents on how to use visual support strategies is recommended.

10.7 Implications of findings for further research on vision assessment and intervention in children with CP

The IQI/VSS approach could be re-investigated in a population exposed to education on PVDas well as the implementation of the visual support strategies for a longer period of time than 6 weeks. The sensitivity and specificity of the objective tests need to investigated in order to identify which are best used in a busy clinic setting and in children who have normal vision with PVD compared with those that have visual impairment and PVD.

Planned educational programmes targeted at the understanding and uptake of visual support strategies to educate parents on CVI/PVD is urgently needed.

There is need to find management strategies for the kind of visual impairment associated withCP which ae effective in this population. study is urgently needed in this population.

10.8 Conclusions

In conclusion, this thesis showed high levels of multiple co-morbidities in this population, including visual impairment. Better tools for assessment and management of these problems are required. Despite identifying key knowledge gaps and areas where further research is needed, this thesis has contributed to understanding of the PVD in children with cerebral palsy and generally visual impairment in these children. Further work is needed to examine the interaction between visual impairment and speech and communication.

11 Reference

1. Lucker-Babel M-F. The right of the child to express views and to be heard: An attempt to interpret Article 12 of the UN Convention on the Rights of the Child. Int'l J Child Rts. 1995;3:391.

2. Dyer SJ. The value of children in African countries–insights from studies on infertility. Journal of Psychosomatic Obstetrics & Gynecology. 2007;28(2):69-77.

3. Nauck B, Klaus D. The varying value of children: Empirical results from eleven societies in Asia, Africa and Europe. Current Sociology. 2007;55(4):487-503.

4. Schieber G, Maeda A. Health Care Financing And Delivery In Developing Countries:Developing countries, which contain 84 percent of the world's population, claim only 11 percent of the world's health spending. Health Affairs. 1999;18(3):193-205.

5. Knafl K, Breitmayer B, Gallo A, Zoeller L. Family response to childhood chronic illness: Description of management styles. Journal of Pediatric Nursing. 1996;11(5):315-26.

6. Whittingham K, Wee D, Sanders MR, Boyd R. Sorrow, coping and resiliency: parentsof children with cerebral palsy share their experiences. Disability and rehabilitation. 2013;35(17):1447-52.

7. Bowman R MD, Law E, Mostyn K, Dutton GN. The 'mirror test' for estimating visualacuity in infants. Br J Ophthalmol. 2010;94(7):882-5.

8. Wanamaker CE, Glenwick DS. Stress, coping, and perceptions of child behavior in parents of preschoolers with cerebral palsy. Rehabilitation Psychology. 1998;43(4):297.

9. Control CfD, Prevention. Economic costs associated with mental retardation, cerebralpalsy, hearing loss, and vision impairment--United States, 2003. MMWR Morbidity and mortality weekly report. 2004;53(3):57.

10. Organization WH. International statistical classification of diseases and related health

problems: World Health Organization; 2004.

11. Organization WH. International classification of functioning, disability and health: ICF: Geneva: World Health Organization; 2001.

12. Simeonsson RJ. ICF-CY: A Universal Tool for Documentation of Disability. Journal of Policy and Practice in Intellectual Disabilities. 2009;6(2):70-2.

13. Rosenbaum P GJ. The 'F-words' in childhood disability: I swear this is how we should think! . Child Care Health Dev. 2012;38(4):457-63.

14. Abdel Malek S RP, Gorter JW. Perspectives on cerebral palsy in Africa: Exploringthe literature through the lens of the International Classification of Functioning, Disabilityand Health. Child Care Health Dev. 2020;46(2):175-86.

15. Majnemer A, Mazer B, editors. New directions in the outcome evaluation of children with cerebral palsy. Seminars in Pediatric Neurology; 2004: Elsevier.

16. Moore LJS, Allegrante JP, Palma M, Lewin J, Carlson MG. Assessment of quality of life needs of children with mild hemiplegic cerebral palsy. Children's Health Care. 2010;39(2):157-71.

17. Tessier DW, Hefner JL, Newmeyer A. Factors related to psychosocial quality of life for children with cerebral palsy. Int J Pediatr. 2014;2014:204386.

18. Liptak GS, O'Donnell M, Conaway M, Chumlea WC, Worley G, Henderson RC, et al. Health status of children with moderate to severe cerebral palsy. Developmental medicine and child neurology. 2001;43(6):364-70.

19. Oeffinger D, Tylkowski C, Rayens M, Davis R, Gorton Iii G, D'Astous J, et al. Gross Motor Function Classification System and outcome tools for assessing ambulatory cerebral palsy: a multicenter study. Developmental medicine and child neurology. 2004;46(5):311-9.

20. Schneider JW, Gurucharri LM, Gutierrez AL, Gaebler-Spira DJ. Health-related quality of life and functional outcome measures for children with cerebral palsy.

Developmental Medicine and Child Neurology. 2001;43(9):601-8.

21. Tsoi WSE, Zhang L, Wang W, Tsang K, Lo SK. Improving quality of life of children with cerebral palsy: a systematic review of clinical trials. Child: care, health and development. 2012;38(1):21-31.

22. Karimi M, Brazier J. Health, health-related quality of life, and quality of life: what is the difference? Pharmacoeconomics. 2016;34(7):645-9.

23. Varni JW BT, Berrin SJ, Sherman SA, Artavia K, Malcarne VL, Chambers HG. The PedsQL in pediatric cerebral palsy: reliability, validity, and sensitivity of the Generic Core Scales and Cerebral Palsy Module. Dev Med Child Neurol. 2006;48(6):442-9.

24. Carlon S, Shields N, Yong K, Gilmore R, Sakzewski L, Boyd R. A systematic reviewof the psychometric properties of Quality of Life measures for school aged children with cerebral palsy. BMC Pediatr. 2010;10:81.

25. Varni JW, Limbers CA, Burwinkle TM. Parent proxy-report of their children's health-related quality of life: an analysis of 13,878 parents' reliability and validity across age subgroups using the PedsQL 4.0 Generic Core Scales. Health Qual Life Outcomes. 2007;5:2.

26. Reddihough DS, Collins KJ. The epidemiology and causes of cerebral palsy. Australian Journal of physiotherapy. 2003;49(1):7-12.

27. Baxter P MC, Goldstein M “The definition and classification of cerebral palsy” Developmental Medicine and Child Neurology. 2007;49:1-44.

28. Rosenbaum P PN, Leviton A, Goldstein M, Bax M, Damiano D, Dan B, Jacobsson B.A report: the definition and classification of cerebral palsy April 2006. Dev Med Child Neurol Suppl. 2007;109:8-14.

29. Kakooza-Mwesige A, Andrews C, Peterson S, Wabwire Mangen F, Eliasson AC, Forssberg H. Prevalence of cerebral palsy in Uganda: a population-based study. The Lancet Global Health. 2017;5(12):e1275-e82.

30. Kakooza-Mwesige A, Forssberg H, Eliasson A-C, Tumwine JK. Cerebral palsy in children in Kampala, Uganda: clinical subtypes, motor function and co-morbidities. BMC research notes. 2015;8(1):166.

31. Gladstone M. A review of the incidence and prevalence, types and aetiology of childhood cerebral palsy in resource-poor settings. Ann Trop Paediatr. 2010;30(3):181-96.

32. Lagunju I OA, Famosaya A. Cerebral palsy in Nigerian children: profile and impact on educational opportunities. DMCN. 2016;58(55).

33. Khandaker G, Muhit M, Karim T, Smithers-Sheedy H, Novak I, Jones C, et al. Epidemiology of cerebral palsy in Bangladesh: a population-based surveillance study. Developmental Medicine & Child Neurology. 2018.

34. Bearden DR, Monokwane B, Khurana E, Baier J, Baranov E, Westmoreland K, et al.

Pediatric Cerebral Palsy in Botswana: Etiology, Outcomes, and Comorbidities. Pediatr Neurol. 2016;59:23-9.

35. Whitley E BJ. Statistics review 4:sample size calculations. Crit Care. 2002;6:335–41.

36. Murthy GV, Mactaggart I, Mohammad M, Islam J, Noe C, Khan AI, et al. Assessing the prevalence of sensory and motor impairments in childhood in Bangladesh using key informants. Arch Dis Child. 2014;99(12):1103-8.

37. Duke R OE, Iso M, Okorie U, Ekwe A, Courtright P, Lewallen S. Using key informants to estimate prevalence of severe visual impairment and blindness in children in Cross River State, Nigeria. J AAPOS. 2013;17(4):381-4.

38. Olusanya BO, Ogunlesi TA, Slusher TM. Why is kernicterus still a major cause of death and disability in low-income and middle-income countries? Archives of disease in childhood. 2014:archdischild-2013-305506.

39. Herrgard E, Luoma L, Tuppuraimen K, Karjalamen S, Martikainen A.

Neurodevelopmental profile at five years of children born at≤ 32 weeks gestation. Developmental Medicine & Child Neurology. 1993;35(12):1083-96.

40. Donald KA, Samia P, Kakooza-Mwesige A, Bearden D. Pediatric cerebral palsy in Africa: a systematic review. Semin Pediatr Neurol. 2014;21(1):30-5.

41. Polack S, Adams M, O'banion D, Baltussen M, Asante S, Kerac M, et al. Children with cerebral palsy in Ghana: malnutrition, feeding challenges, and caregiver quality of life. Developmental Medicine & Child Neurology. 2018.

42. Group. SC. Surveillance of cerebral palsy in Europe: a collaboration of cerebral palsysurveys and registers. Developmental Medicine and Child Neurology. 2000;42:816-24.

43. Liptak GS. Overview of GMFCS.

44. Palisano R, Rosenbaum P, Walter S, Russell D, Wood E, Galuppi B. Developmentand reliability of a system to classify gross motor function in children with cerebral palsy. Developmental Medicine & Child Neurology. 1997;39(4):214-23.

45. Gunel K M MA, Tarsuslu Tulay, Livanelioglu A. Relationship among the Manual Ability Classification System (MACS), the Gross Motor Function Classification System (GMFCS), and the functional status (WeeFIM) in children with spastic cerebral palsy. Eur J Pediatr 2009;168:477-85.

46. Clarke M, Newton C, Griffiths T, Price K, Lysley A, Petrides K. Factors associated with the participation of children with complex communication needs. Research in developmental disabilities. 2011;32(2):774-80.

47. Voorman JM, Dallmeijer AJ, Van Eck M, Schuengel C, Becher JG. Social functioning and communication in children with cerebral palsy: association with disease characteristics and personal and environmental factors. Developmental Medicine & Child Neurology. 2010;52(5):441-7.

48. Virella D, Pennington L, Andersen GL, Andrada MdG, Greitane A, Himmelmann K,

et al. Classification systems of communication for use in epidemiological surveillance of children with cerebral palsy. Developmental Medicine & Child Neurology. 2016;58(3):285-91.

49. Dormans JP, Pellegrino L. Caring for Children with Cerebral Palsy: A Team Approach: ERIC; 1998.

50. Donald KA, Kakooza AM, Wammanda RD, Mallewa M, Samia P, Babakir H, et al. Pediatric cerebral palsy in Africa: where are we? Journal of child neurology. 2015;30(8):963-71.

51. Green SE. "What do you mean 'what's wrong with her?'": stigma and the lives of families of children with disabilities. Social Science & Medicine. 2003;57(8):1361-74.

52. Aran A. Quality of Life in Children with Cerebral Palsy. In: Preedy VR, Watson RR, editors. Handbook of Disease Burdens and Quality of Life Measures. New York, NY: Springer New York; 2010. p. 2453-68.

53. WHO. Universal eye health:a global action plan 2014-2019. World Health Organization. 2013.

54. Fiona Gorrie KG, Robert Rush,John Ravenscroft,. Towards population screening for Cerebral Visual Impairment: Validity of the Five Questions and the CVI Questionnaire. PLoSOne. 2019;14(3):e0214290.

55. Gogate P, Kalua K, Courtright P. Blindness in childhood in developing countries: time for a reassessment? PLoS Med. 2009;6(12):e1000177.

56. Gilbert C, Muhit M. Eye conditions and blindness in children: priorities for research, programs, and policy with a focus on childhood cataract. Indian J Ophthalmol. 2012;60(5):451-5.

57. Rahi JS, Cable N, Group BCVIS. Severe visual impairment and blindness in children in the UK. The Lancet. 2003;362(9393):1359-65.

58. Blencowe H, Lawn JE, Vazquez T, Fielder A, Gilbert C. Preterm-associated visual impairment and estimates of retinopathy of prematurity at regional and global levels for2010. Pediatric research. 2013;74(S1):35.

59. O''Connor R Anna Terence Stephson AJ, Michael J Tobin, Merrick J Moseley, Sonia Ratib, Alistair Fielder. Long-Term Ophthalmic Outcome of Low Birth Weight Children With and Without Retinopathy of Prematurity. PEDIATRICS. 2012;109(1):12-7.

60. Ego A LK, Brovedani P, Belmonti V, Gonzalez-Monge S, Boudia B, Ritz A, Cans C. Visual-perceptual impairment in children with cerebral palsy: a systematic review. Dev Med Child Neurol. 2015;57(2):46-51.

61. Good WV, Jan JE, Burden SK, Skoczenski A, Candy R. Recent advances in cortical visual impairment. Developmental medicine and child neurology. 2001;43(1):56-60.

62. Boot FH, Pel JJ, van der Steen J, Evenhuis HM. Cerebral Visual Impairment: which perceptive visual dysfunctions can be expected in children with brain damage? A systematic review. Res Dev Disabil. 2010;31(6):1149-59.

63. Sakki HEA, Dale NJ, Sargent J, Perez-Roche T, Bowman R. Is there consensus in defining childhood cerebral visual impairment? A systematic review of terminology and definitions. Br J Ophthalmol. 2018;102(4):424-32.

64. Dutton G BJ, Boyd G, Bradnam M, Day R, McCulloch D, Mackie R, Phillips S, Saunders K. Cortical visual dysfunction in children: A clinical study Eye. 1996;10:302-9.

65. Ahmed M, Dutton G. Cognitive visual dysfunction in a child with cerebral damage. Developmental Medicine & Child Neurology. 1996;38(8):736-9.

66. Josef Zihl GND. Visuoperceptive and Visuocognitive Disorders. Cerebral Visual Impairment in Children: Springer-Verlag Wien; 2015. p. 11-50.

67. Jan J, Groenveld M, Sykanda A, Hoyt C. Behavioural characteristics of children with permanent cortical visual impairment. Developmental medicine & child neurology.

1987;29(5):571-6.

68. Philip SS, Dutton GN. Identifying and characterising cerebral visual impairment in children: a review. Clin Exp Optom. 2014;97(3):196-208.

69. GN D. Cognitive vision, its disorders and differential diagnosis in adults and children:knowing where and what things are. Eye. 2003;17(17):289-304.

70. Goldstein EB. Introduction to perception. Sensation and perception: Wadsworth; 2010. p. 3-20.

71. Goodale MA. Separate visual systems for perception and action: a framework for understanding cortical visual impairment. Developmental Medicine & Child Neurology. 2013;55.

72. Sonksen PM DN. Visual impairment in infancy: impact on neurodevelopmental and neurobiological processes. Arch Dis Child 2. 2014;99:1163-8.

73. Braddick O, Atkinson J. Development of human visual function. Vision Res. 2011;51(13):1588-609.

74. Dutton GN, Jacobson LK. Cerebral visual impairment in children. Semin Neonatol. 2001;6(6):477-85.

75. Ortibus E LA, Verhoeven J, De Cock P, Casteels I, Schoolmeesters B, Buyck A, Lagae L. Screening for cerebral visual impairment: value of a CVI questionnaire. Neuropediatrics 2011;42:138-47.

76. Good WV. Development of a quantitative method to measure vision in children with chronic cortical visual impairment Tr Am Ophth Soc. 2001;99:253-67.

77. Zaporozhets AV. The development of perception in the preschool child. Monographs of the Society for Research in Child Development. 1965;30(2):82-101.

78. Macintyre-Beon C IH, Hay I, Cockburn D, Calvert J, Dutton G. Dorsal Stream dysfunction in Children: A Review and an Approach to Diagnosis and Management. Current

Pediatric Reviews 2010;6:166-82.

79. Dutton GN Saeed A Fahad B. The association of binocular lower visual field impairment, impaired simultaneous perception, disordered visually guided motion and inaccurate saccades in children with cerebral visual dysfunction – a retrospective observational study. Eye. 2004;18:27–34.

80. Dutton GN. Cognitive visual dysfunction. The British journal of ophthalmology.

1994;78(9):723.

81. Dutton G, Bax M. Visual impairment in children due to damage to the brain: Clinics in Developmental Medicine: John Wiley & Sons; 2010.

82. CS H. Visual function in the brain-damaged child. Eye 2003;17(3):369-84.

83. Chokron S, Dutton GN. Impact of Cerebral Visual Impairments on Motor Skills: Implications for Developmental Coordination Disorders. Front Psychol. 2016;7:1471.

84. Sakki H DN, Sargent J, Perez-Roche T, Bowman R. Is there consensus in defining childhood cerebral visual impairment? A systematic review of terminology and definitions. British Journal of Ophthalmology 2018;102:424-32.

85. Huo R, Burden SK, Hoyt CS, Good WV. Chronic cortical visual impairment in children:

aetiology, prognosis, and associated neurologicaldeficits. Br J Ophthalmol. 1999;83:670-5.

86. Stiers P VR, Vanneste G, Coene S, De Rammelaere M, Vandenbussche E. Visual–perceptualimpairment in arandom sample ofchildren with cerebralpalsy. Developmental Medicine & Child Neurology. 2002;44:370.

87. McCulloch DL MR, Dutton GN, Bradnam MS, Day RE, McDaid GJ , Phillips S, Napier A, Herbert A M, Saunders KJ , Shepherd AJ A visual skills inventory for children with neurological impairments. Developmental Medicine & Child Neurology. 2007;49:757–63

88. Gordon Dutton MB. Impairment of cognitive vision: its detection and measurement.

Visual Impairment in Children due to Damage to the Brain. 2010:224.

89. Ferziger NB, Nemet P, Brezner A, Feldman R, Galili G, Zivotofsky AZ. Visual assessment in children with cerebral palsy: implementation of a functional questionnaire. DevMed Child Neurol. 2011;53(5):422-8.

90. Dutton GN. Structured history taking to characterize visual dysfunction and plan optimal habilitation for children with cerebral visual impairment. Dev Med Child Neurol. 2011;53(5):390.

91. Mitry D WC, Northstone K, Akter A, Jewel J, Khan N, Muhit M, Gilbert C, Bowman

R. Perceptual visual dysfunction, physical impairment and quality of life in Bangladeshi children with cerebral palsy. British Journal of Ophthlamology. 2016;100(0):1245-50.

92. Tsirka A Liasis A, Kuczynski A, Vargha-

Khadem F, Kukadia R, Dutton G, Bowman R. Clinical use of the Insight Inventory in cerebral visual impairment and the effectiveness of tailored habilitational strategies Dev Med& Child Neurol 2020;62(11):1324-30.

93. Novak I, Mcintyre S, Morgan C, Campbell L, Dark L, Morton N, et al. A systematic review of interventions for children with cerebral palsy: state of the evidence. DevelopmentalMedicine & Child Neurology. 2013;55(10):885-910.

94. Philip SS TS, Thomas MM, Dutton GN, Bowman R. A Validation of an Examination Protocol for Cerebral Visual Impairment Among Children in a Clinical Population in India. J Clin Diagn Res 2016 10(12):01-4.

95. Duke RE NK, Torty C, et al. Visual impairment and perceptual visual disorders in children with cerebral palsy in Nigeria. Br J Ophthalmol. 2020; **0:1**–8

96. Macintyre-Beon C, Young D, Calvert J, Ibrahim H, Dutton GN, Bowman R. Reliability of a question inventory for structured history taking in children with cerebral

visual impairment. Eye (Lond). 2012;26(10):1393.

97. Philip SS. Setting up of a cerebral visual impairment clinic for children: Challenges and future developments. Indian Journal of Ophthalmology. 2017;65:30-4.

98. Vani JW Said M KP. The PedsQL in pediatric cerebral palsy: reliability, validity, andsensitivity of the Generic Core Scales and Cerebral Palsy Module. Developmental Medicine & Child Neurology 2006;48:442–449 2006;48:442-9.

99. Viehweger E, Robitail S, Rohon MA, Jacquemier M, Jouve JL, Bollini G, et al.

Measuring quality of life in cerebral palsy children. Annales de Réadaptation et de Médecine Physique. 2008;51(2):129-37.

100. White-Koning M, Arnaud C, Dickinson HO, Thyen U, Beckung E, Fauconnier J, et al. Determinants of child-parent agreement in quality-of-life reports: a European study of children with cerebral palsy. Pediatrics. 2007;120(4):e804-14.

101. Macintyre-Béon C, Young D, Dutton GN, Mitchell K, Simpson J, Loffler G, et al.

Cerebral visual dysfunction in prematurely born children attending mainstream school. Documenta Ophthalmologica. 2013;127(2):89-102.

102. Christopher R. Bennett PJB, Corinna M. Bauer, and Lotfi B. Merabet. The Assessment of Visual Function and Functional Vision. Semin Pediatr Neurol. 2019;31:30-40.

103. King S, Teplicky R, King G, Rosenbaum P, editors. Family-centered service for children with cerebral palsy and their families: a review of the literature. Seminars in pediatric neurology; 2004: Elsevier.

104. Fatemeh Rajati HA, Nader Salari, Masood Ghanbari, Zahra Naghibifar, Seyed Younes Hosseini. Quality of life predictors in physically disabled people. J Educ Health Promot. 2018;7(61):115-7.

105. Huber M, Knottnerus, J. A., Green, L., van der Horst, H., Jadad, A. R., Kromhout, D.,
Leonard, B., Lorig, K., Loureiro, M. I., van der Meer, J. W., Schnabel, P., Smith, R., van Weel, C. Smid, H. How should we define health? BMJ (Clinical Research Ed). 2011;343(d4163).

106. Majnemer A, Shevell M, Rosenbaum P, Law M, Poulin C. Determinants of life quality in school-age children with cerebral palsy. J Pediatr. 2007;151(5):470-5, 5 e1-3.

107. Heather O Dickinson KNP, Ulrike Ravens-Sieberer, Giorgio Schirripa, Ute Thyen, Catherine Arnaud, Eva Beckung, Jérôme Fauconnier, Vicki McManus, Susan I Michelsen, Jackie Parkes, Allan F Colver. Self-reported quality of life of 8–12-year-old children with cerebral palsy: a cross-sectional European study. Lancet. 2007;369:2171-78.

108. Lagunju I OA, Akinyinka O, Akinbami F. Health-related Quality of life of Nigerian children with epilepsy. African Journal of Neurological Sciences. 2009;28(1):23-36.

109. Macro I, Commission NP. Nigeria demographic and health survey 2013. 2014.

110. Hambleton RK. The next generation of the ITC Test Translation and Adaptation Guidelines. European Journal of Psychological Assessment. 2001;17(3):164.

111. Scientific L. What is Lingusitic Validation: Language Sientific; 2020. Available from: https://www.languagescientific.com/what-is-linguistic-validation/.

112. Raina P ODM, Rosenbaum P, Brehaut J, Walter S, Russell D, Swinton M, Zhu B. TheHealth and Well-Being of Caregivers of Children With Cerebral Palsy. Pediatrics 2005;115(6):e626-36.

113. Winter S, Autry A, Boyle C, Yeargin-Allsopp M. Trends in the Prevalence of Cerebral Palsy in a Population-Based Study. Pediatrics 2002;110:1220-5.

114. Shevell MI, Dagenais L, Hall N, Consortium R. Comorbidities in cerebral palsy and their relationship to neurologic subtype and GMFCS level. Neurology. 2009;72(24):2090-6.

115. WC C. Providing a primary care medical home for children and youth with

cerebral.

Pediatrics. 2004;114:1106-13.

116. O'Shea M. Cerebral palsy. Semin Perinatol. 2008;32(1):35-41.

117. Nottidge VA OM. Cerebral palsy in Ibadan Nigeria. Dev Med Child Neurol 1991;33(241-5).

118. Belonwu R GG, Adeleke S. Cerebral palsy in Dar Es Salaam. Cent Afr J Med. 1990 36:8-10.

119. MH. J. Functions in brain development in human. Nat Rev Neurosci. 2001;45(1566-72).

120. Cole M CS. The development of children. New York: Worth Publ; 2001.

121. Goodman R. The Strengths and Difficulties Questionnaire: A Research Note. J Child Psychol Psychiat. 1997;35(5):581-6.

122. Fazzi E, Signorini SG, R LAP, Bertone C, Misefari W, Galli J, et al. Neuro-ophthalmological disorders in cerebral palsy: ophthalmological, oculomotor, and visual aspects. Dev Med Child Neurol. 2012;54(8):730-6.

123. Dutton GN. 'Dorsal stream dysfunction' and 'dorsal stream dysfunction plus':a potential classification for perceptual visual impairment in thecontext of cerebral visual impairment? Developmental Medicine & Child Neurology 2009;51:168-72.

124. Williams C, Northstone K, Sabates R, Feinstein L, Emond A, Dutton GN. Visual perceptual difficulties and under-achievement at school in a large community-based sample of children. PLoS One. 2011;6(3):e14772.

125. A G, E M, G. C. Visual disorders in children with brain lesions: 2. Visual impairmentassociated with cerebral palsy. Eur J Paediatr Neurol 2001;5:115-9.

126. McCullough N, Parkes J, Kerr C, McDowell BC. The health of children and young people with cerebral palsy: a longitudinal, population-based study. Int J Nurs Stud. 2013;50(6):747-56.

127. Houliston MJ TA, Dutton GN, Young DG. Evidence of cognitive visual problems in children with hydrocephalus: a structured clinical history- taking strategy. Dev Med Child Neurol. 1999;41:298-306.

128. chasm. CoQoHCiAIoMCtq. A new health system for the 21st century. Washington, DC: National Academy Press; 2001.

129. Ellenberg JH, Nelson KB. The association of cerebral palsy with birth asphyxia: a definitional quagmire. Dev Med Child Neurol. 2013;55(3):210-6.

130. Van den Bos AMG TH. Quality of life as an instrument for need 834 assessment and outcome assessment of health care in chronic patients. Quality in Health Care. 1999(8247-52).

131. J V. Scaling and Scoring of the Pediatric Quality of Life Inventory PedsQL. 2015.

132. Pansell T, Hellgren K, Jacobson L, Brautaset R, Tedroff K. The accommodative process in children with cerebral palsy: different strategies to obtain clear vision at short distance. Developmental Medicine & Child Neurology. 2014;56(2):171-7.

133. Commission P. Cross River State clan and population edict. 2007.

134. Muniz J, Hambleton RK, Xing D. Small sample studies to detect flaws in item translations. International Journal of Testing. 2001;1(2):115-35.

135. Fonteyn ME, Kuipers B, Grobe SJ. A Description of Think Aloud Method and Protocol Analysis. Qualitative Health Research. 1993;1.

136. Lundgrén-Laine H, Salanterä S. Think-Aloud Technique and Protocol Analysis in Clinical Decision-Making Research. Qualitatitve Health Research [Internet]. 2009 18th June 2018; 3. Available from: http://journals.sagepub.com/doi/pdf/10.1177/1049732309354278.

137. Ward L, DeanTraweek. Application of a metacognitive strategy to assessment, intervention, and consultation: A think-aloud technique. Journal of School Psychology.31(4):469-85.

138. Andrews C, Kakooza-Mwesige A, Elisson A-C, Forssberg H. Important report on
cerebral palsy in Bangladesh: but different findings compared with other countries need further exploration. Developmental Medicine & Child Neurology. 2019;61(5):511-2.

139. Peter Rosenbaum NP, Alan Leviton, Murray Goldstein, and Martin Bax. The Definition and Classification of Cerebral Palsy. Dev Med Child Neurol. 2007;49:(Suppl. 109): 8–14.

140. Smithers-Sheedy H, Badawi N, Blair E, Cans C, Himmelmann K, Krägeloh-Mann I, et al. What constitutes cerebral palsy in the twenty-first century? Developmental Medicine & Child Neurology. 2014;56(4):323-8.

141. Himpens E, Van den Broeck C, Oostra A, Calders P, Vanhaesebrouck P. Prevalence, type, distribution, and severity of cerebral palsy in relation to gestational age: a meta-analyticreview. Developmental Medicine & Child Neurology. 2008;50(5):334-40.

142. Pakula AT, Van Naarden Braun K, Yeargin-Allsopp M. Cerebral palsy: classification and epidemiology. Phys Med Rehabil Clin N Am. 2009;20(3):425-52.

143. Blencowe H, Vos T, Lee AC, Philips R, Lozano R, Alvarado MR, et al. Estimates of neonatal morbidities and disabilities at regional and global levels for 2010: introduction, methods overview, and relevant findings from the Global Burden of Disease study. Pediatr Res. 2013;74 Suppl 1:4-16.

144. National Malaria Elimination Programme (NMEP) NPCN, National Bureau of Statistics (NBS), and ICF International. Nigeria Malaria Indicator. Abuja, Nigeria, and Rockville, Maryland, USA: NMEP, NPopC, and ICF International. ; 2015.

145. Mung'ala-Odera V MR, Njuguna P, Mturi N, Alcock K, Carter JA, Newton CR. Validity and reliability of the 'Ten Questions' questionnaire for detecting moderate to severe neurological impairment in children aged 6-9 years in rural Kenya. Neuroepidemiology.23(1-2):67-72.

146. Gilstrap JETLC. Help for perinatal prevention of cerebral palsy. JAMA. 2003;290(20):2730-2.

147. Astrid Nystedt IH. Diverse definitions of prolonged labour and its consequences with sometimes subsequent inappropriate treatment. BMC Pregnancy and Childbirth. 2014;14:233.

148. Lambert SR. Congenital rubella syndrome: the end is in sight. BMJ Publishing GroupLtd; 2007.

149. Orenstein WA, Preblud SR, Bart KJ, Hinman AR. Methods of assessing the impact ofcongenital rubella infection. Clinical Infectious Diseases. 1985;7(Supplement_1):S22-S8.

150. Olusanya BO, Osibanjo FB, Slusher TM. Risk factors for severe neonatal hyperbilirubinemia in low and middle-income countries: a systematic review and meta-analysis. PLoS One. 2015;10(2):e0117229.

151. Blair E, Stanley FJ. Intrapartum asphyxia: a rare cause of cerebral palsy. The Journal of pediatrics. 1988;112(4):515-9.

152. Stavsky M, Mor O, Mastrolia SA, Greenbaum S, Than NG, Erez O. Cerebral Palsy—Trends in Epidemiology and Recent Development in Prenatal Mechanisms of Disease, Treatment, and Prevention. Frontiers in pediatrics. 2017;5:21.

153. Robertson S, Cutts F, Samuel R, Diaz-Ortega J. Control of rubella and congenital rubella syndrome (CRS) in developing countries, Part 2: Vaccination against rubella. Bulletinof the World Health Organization. 1997;75(1):69.

154. Vargus-Adams J. Health-related quality of life in childhood cerebral palsy. Archives of physical medicine and rehabilitation. 2005;86(5):940-5.

155. Chataika T, McKenzie JA, Swart E, Lyner-Cleophas M. Access to education in Africa: responding to the United Nations Convention on the Rights of Persons with Disabilities. Disability & Society. 2012;27(3):385-98.

156. Aron L, Loprest P. Disability and the Education System. The Future of

Children.
2012;22(1):97-122.

157. Watson SF. Barriers to inclusive education in Ireland: The case for pupils with a diagnosis of intellectual and/or pervasive developmental disabilities. British Journal of Learning Disabilities. 2009;37(4):277-84.

158. Rosenbaum P, Stewart D. The world health organization international classification of functioning, disability, and health: a model to guide clinical thinking, practice and research in the field of cerebral palsy. Seminars in Pediatric Neurology. 2004;11(1):5-10.

159. Bank W. NIGERIA-Socio Economic Assessment. Washington, DC: The World Bank; 2011. Contract No.: 70606.

160. Education FMO. Nigeria Digest of Education Statistics Nigeria2020. [https://education.gov.ng/nigeria-digest-of-education-statistics/].

161. report Gn. Global Nutrition report: Nigeria 2020 [Available from: https://globalnutritionreport.org/resources/nutrition-profiles/africa/western-africa/nigeria/.

162. Duke R TC, Nwachukwu K, Ameh S, Kim N, Eneli N, Onyedikachi A, Aghaji A, Burton K, Dyet L, Bowman R. Clinical features and aetiology of cerebral palsy in children from Cross River State, Nigeria. Arch Dis Child 2020;0:1-6.

163. Duke R EK, Burton K, MacLeoad D,Dutton GN, Gilbert C, Bowman R. The effect ofvisual support strategies on the quality of life of children with cerebral palsy and cerebral visual impairment/ perceptual visual dysfunction in Nigeria: study protocol for a randomized controlled trial. Trials. 2019;20:417.

164. Ndem Ayara UE, Enang Udah. The universalization of basic education in Nigeria: theCross River state experience. Wudpecker Journal of Public Administration 2013;1(1):007-19.

165. Adamu Sa'idu Adamu UAS, Garba Dayyabu Gwarzo, Raymond O Belonwu. Nutritional status in cerebral palsy: A Cross-sectional comparative survey of children in

Kano, Nigeria. Nigerian Post Graduate Medical Journal. 2018;25(3):156-60.

166. Mona P. Gajre VD, Rashmi Yeradkar,Arpita Adhikari. Study of visual perception problems in children with learning disability. ndian Journal of Basic and Applied Medical Research;. 2015;4(3):492-7.

167. Nesbitt RC MS, Kuper H, Muhit M, Murthy GV. Predictors of referral uptake in children with disabilities in Bangladesh – Exploring barriers as a first step to improvingreferral provision. Disability and Rehabilitation. 2011;34(13):1089-95.

168. Sue Mackey GVSM, Mohammad A. Muhit, Johurul J. Islam, and Allen Foster. Validation of the Key Informant Method to IdentifyChildren with Disabilities: Methods and Results from a Pilot Study in Bangladesh. Journal of Tropical Pediatrics. 2012;58(4):269-74.

169. Muhit MA, Shah SP, Gilbert CE, Hartley SD, Foster A. The key informant method: a novel means of ascertaining blind children in Bangladesh. Br J Ophthalmol. 2007;91(8):995-9.

170. Palisano R, Rosenbaum P, Walter S, Russel D, Wood E, Galuppi B. Development andreliability of a system to classify gross motor function in chldren with cerebral palsy. Developmental medicne and child neurology. 1997;39:214-23.

171. Khandaker G, Smithers-Sheedy H, Islam J, Alam M, Jung J, Novak I, et al. Bangladesh Cerebral Palsy Register (BCPR): a pilot study to develop a national cerebral palsy (CP) register with surveillance of children for CP. BMC Neurol. 2015;15:173.

172. Francesca Bertuzzi JGO, 2 Maria Rita Porta, Gian Paolo Paliaga, Stefano Miglior.

Sensitivity and specificity of a visual acuity screening protocol performed with the Lea Symbols 15-line folding distance chart in preschool children. Acta Ophthalmologica Scandinavica. 2006;84:807–11.

173. Scruggs TE, Mastropieri MA. On babies and bathwater: Addressing the problems of identification of learning disabilities. Learning Disability Quarterly. 2002;25(3):155-68.

174. Thomas K GAU, Shekhar P S. Clinical Practice Guidelines for Assessment and
Management of intellectual disability. Indian J Psychiatry. 2019;61(2):194-210.

175. Strand EA, McCauley RJ. Differential Diagnosis of Severe Speech Impairment in Young Children.

176. Sander J SS. Incidence and prevalence studies in epilepsy and their methodological problems: a review. Journal of Neurology, Neurosurgery, and Psychiatry 1987;50:829-39.

177. Westergren A LC, Axelsson C, Ulander K. Prevalence of eating difficulties and malnutrition among persons within hospital care and special accommodations. The Journal ofNutrition Health and Aging. 2008;12(1):39-43.

178. Gunel K M MA TT, Livanelioglu A. Relationship among the Manual Ability Classification System (MACS), the Gross Motor Function Classification System (GMFCS), and the functional status (WeeFIM) in children with spastic cerebral palsy. Eur J Pediatr . 2009;168:477-85.

179. Nikhat Hoosen ELD, Petrus J. de Vries,Maylene Shung-King. The Strengths and Difficulties Questionnaire (SDQ) in Africa: a scoping review of its application and validation. Child Adolesc Psychiatry Ment Health. 2018;12(6).

180. Van der Meer M, Dixon A, Rose D. Parent and child agreement on reports of problembehaviour obtained from a screening questionnaire, the SDQ. European child & adolescent psychiatry. 2008;17(8):491-7.

181. Schlenker J RS. A health professional's guide to using growth charts. Paediatr Child Health. 2004;9(3).

182. Kuczmarski R.J OCL, Grummer-Strawn L.M, Flegal K.M, Guo S.S, Wei R CDC growth charts: United States. Adv Data. 2000;314:1-27.

183. Organization WH, Unicef. WHO child growth standards and the identification of severe acute malnutrition in infants and children: joint statement by the World Health

Organization and the United Nations Children's Fund. 2009.

184. Wichers MJ, Odding E, Stam H, van Nieuwenhuizen O. Clinical presentation, associated disorders and aetiological moments in Cerebral Palsy: a Dutch population-based study. Disability and rehabilitation. 2005;27(10):583-9.

185. Michael I. Shevell LD, Nicholas Hall, and On behalf of the REPACQ Consortium.

Comorbidities in cerebral palsy and their relationship to neurologic subtype and GMFCS level. Neurology. 2009;72(24):2090-6.

186. Gangil A PA, Aneja S, Ahuja B, Anand VK. Feeding problems in children with cerebral palsy. Indian Pediatr. 2001;38(3):839-46.

187. JC A. Feeding children with cerebral palsy and swallowing difficulties. European Journal of Clinical Nutrition 2013;67:S9-S12.

188. Valderas JM, Starfield B, Sibbald B, Salisbury C, Roland M. Defining comorbidity: implications for understanding health and health services. Ann Fam Med. 2009;7(4):357-63.

189. Aldenkamp AP, Weber B, Overweg-Plandsoen WC, Reijs R, van Mil S. Educational underachievement in children with epilepsy: a model to predict the effects of epilepsy on educational achievement. Journal of Child Neurology. 2005;20(3):175-80.

190. Saunders K J MJF. Spectacle intervention in children with cerebral palsy (CP) and accommodative dysfunction. Investigative Ophthalmology & Visual Science 2004;45(13).

191. Aneja S AB, Taluja V, Bhatia VK. Epilepsy in children with cerebral palsy. Indian JPediatr. 2001;68(2):111-5.

192. Odding E, Roebroeck ME, Stam HJ. The epidemiology of cerebral palsy: incidence, impairments and risk factors. Disabil Rehabil. 2006;28(4):183-91.

193. Fazzi E, Signorini SG, Bova SM, La Piana R, Ondei P, Bertone C, et al. Spectrum ofvisual disorders in children with cerebral visual impairment. Journal of child neurology. 2007;22(3):294-301.

194. Hyvarinen L. LEA Vision Test System for Assessment and Screening Elgin,IL: Good-Lite; [Available from: www.lea-test.fi.

195. David McShefferty WMW, Iain R C Swan, Michael A Akeroyd. The effect ofexperience on the sensitivity and specificity of the whispered voice test: a diagnostic accuracy study. BMJ Open. 2012;3(4).

196. Ghasia F, Brunstrom J, Gordon M, Tychsen L. Frequency and severity of visual sensory and motor deficits in children with cerebral palsy: gross motor function classificationscale. Invest Ophthalmol Vis Sci. 2008;49(2):572-80.

197. Pilling RF OL, Bruce A. Assessing visual function in children with complex disabilities: the Bradford visual function box. Br J Ophthalmol. 2016;100(8):1118-21.

198. Fazzi E, Signorini SG, La Piana R, Bertone C, Misefari W, Galli J, et al. Neuro-ophthalmological disorders in cerebral palsy: ophthalmological, oculomotor, and visual aspects. Developmental Medicine & Child Neurology. 2012;54(8):730-6.

199. Ghasia F, Brunstrom-Hernandez J, Tychsen L. Repair of strabismus and binocular fusion in children with cerebral palsy: Gross motor function classification scale. Investigativeophthalmology & visual science. 2011;52(10):7664-71.

200. Tychsen L. The Cause of Infantile Strabismus Lies Upstairs in the Cerebral Cortex,Not Downstairs in the Brainstem. Arch Ophthalmol. 2012;130(8):1060-1.

201. Brodsky MC. Motion Responses in Human Strabismus: What Optokinesis in the Deviating Eye Is Telling Us. Investigative Ophthalmology & Visual Science. 2016;57:2990.

202. Saunders JFMJPNHAJJKJ. Accommodative Dysfunction in Children with CerebralPalsy: A Population-Based Study. Investigative Ophthalmology & Visual Science. 2006;47:1824-30.

203. Swetha Sara Philip ST, Maya Mary Thomas,Gordon N Dutton, Richard Bowman. A Validation of an Examination Protocol for Cerebral Visual Impairment Among Children in a Clinical Population in India. J Clin Diagn Res. 2016;10(2):NC01-NC4.

204. Bodunde O. T A-PD, Ojuawo A, Adeboye M.A.N. Ocular findings in children with cerebral

palsy attending a tertiary hospital in North Central Nigeria. Sierra Leone Journal ofBiomedical Research. 2015;7(2):1-7.

205. Pehere N, Chougule P, Dutton GN. Cerebral visual impairment in children: Causes and associated ophthalmological problems. Indian Journal of Ophthalmology. 2018;66:812-5.

206. Becker R HS, Gräf H M, Kaufmann H. Examination of young children with Leasymbols. Br J Ophthalmol. 2002;86:513-6.

207. Saunders KJ, McClelland JF, Richardson PM, Stevenson M. Clinical judgement of near pupil responses provides a useful indicator of focusing ability in children with cerebralpalsy. Dev Med Child Neurol. 2008;50(1):33-7.

208. Saunders KJ, Little JA, McClelland JF, Jackson AJ. Profile of refractive errors incerebral palsy: impact of severity of motor impairment (GMFCS) and CP subtype on refractive outcome. Invest Ophthalmol Vis Sci. 2010;51(6):2885-90.

209. Williams C, Gilchrist ID, Fraser S, McCarthy HM, Parker J, Warnes P, et al. Normative data for three tests of visuocognitive function in primary school children: cross-sectional study. Br J Ophthalmol. 2015;99(6):752-6.

210. Surman G, Bonellie S, Chalmers J, Colver A, Dolk H, Hemming K, et al. UKCP: a collaborative network of cerebral palsy registers in the United Kingdom. J Public Health (Oxf). 2006;28(2):148-56.

211. Pennington L, Goldbart J, Marshall J. Speech and language therapy to improve the communication skills of children with cerebral palsy. Cochrane Database of Systematic Reviews. 2004(2).

212. Belonwu R O GGD, Adeleke S I. Cerebral Palsy in Kano, Nigeria - A Review. Nigerian Journal of Medicine. 2009;18(2):186-9.

213. Aghaji A, Okoye O, Bowman R. Causes and emerging trends of childhood blindness: findings from schools for the blind in Southeast Nigeria. Br J Ophthalmol. 2015;99(6):727-31.

214. Olusanya BO OT, Kumar P, Boo NY, Iskander IF, de Almeida MF, et al.

Management of late-preterm and term infants with hyperbilirubinaemia inresource-constrained settings. BMC Pediatr 15-39.

12 Appendix -Questionnaires and survey tools

Appendix 12.1: English version of the Insight Questions Inventory

About your child	
Child's forename:	Surname:
D.O.B.:	Gender:
Instructions to parents	
These questions are designed for a range of ages, so some questions may seem odd. Your child may have difficulty with some behaviours listed below but not others – this is normal.	
For each of the items listed, please could you click on the box next to it and select which best fits with your child's present behaviour: never/ rarely/ sometimes/ often/ always/ not applicable (NA).	
Questions	Responses
1 Does your child trip or bump into things on the floor?	

2	Does your child find it hard to walk down stairs?			
3	Does your child trip or seem not to notice when pavements go up or down?			
4	Does your child seem to ‘get stuck’ at the top of a slide or hill?			
5	Does your child look down when they cross floor boundaries, for example where lino meets carpet?			
6	Does your child leave food on their plate? If so, is this on the near or far side?			
7	Does your child leave food on their plate? If so, is this on the left or right side?			
8	Does your child have difficulty stepping into the bath, apart from any problems with balance?			
9	Does your child find it hard to find the beginning of a line or the next word when reading, or miss pictures or words on one side of a page (left /right)?			
10	Does your child walk ,move out in front of traffic (coming from left/ right / both)?			
11	Does your child bump into door frames or partly open doors (left/right/both)?			
12	Does your child have difficulty seeing things from a moving vehicle?			

13	Does your child find it hard to spot things that are moving quickly, such as children, small animals?			
14	Does your child find it hard to see moving water i.e. to fill a cup?			
15	Does your child find it hard to follow a cursor on the computer screen?			
16	Does your child avoid watching fast moving TV programmes, and prefer to watch slow moving TV programmes?			
17	Does your child find it hard to catch a ball?			
18	When walking, does your child hold onto your clothes, tugging down?			
19	Does your child find uneven ground hard to walk over?			
20	Does your child bump into low furniture, for example a small table?			
21	Does your child get angry if furniture is moved?			
22	Does your child explore floor boundaries (for example, where lino meets a carpet) with their foot before crossing it or find floor boundaries difficult to cross?			

23	Does your child reach incorrectly for objects (i.e. reaches beyond or around the object or when picking it up, grasp incorrectly, missing or knocking the object over)?			
24	Does your child find it hard to spot something pointed out in the distance?			
25	Does your child find it hard to spot a close friend or relative standing in a group?			
26	Does your child have difficulty playing team games with a lot of players, especially if there are a lot of players and everyone is moving?			
27	Does your child have difficulty finding an item on supermarket or cupboard shelf e.g. finding the cereal or biscuits?			
28	Does your child find it hard to spot an item of clothing in a pile of clothes?			
29	Does your child find it hard to spot the toy they want in a full toy box, shoe in a box of shoes etc?			
30	Does your child find it hard to spot objects when they are on a similar background, such as a white tee shirt on a white sheet?			
31	Does your child get lost in places where there is a lot to see, e.g. in a crowded shopping centre?			

32	Does your child find copying words or drawings time consuming and difficult?			
33	Does your child have difficulty reading crowded text on paper or on a computer screen, but can cope better if some of the text is covered or taken away?			
34	Does your child find it hard to spot letters on a keyboard but knows the alphabet?			
35	Does your child sit closer to the television than about 30 cm?			
36	Does your child find it difficult to keep to task for more than 5 minutes, or after being distracted do they find it difficult to get back to what they were doing?			
37	Does your child react angrily when other children cause distractions?			
38	Does your child bump into things when they are walking and talking at the same time?			
39	Does your child miss objects that are obvious to you because they are different from their background and seem to ‘pop out’ e.g. a bright ball in the grass?			
40	Does your child get upset in busy places, such as supermarkets or shopping centres?			

41	Does your child find it difficult to recognise close relatives in real life?			
42	Does your child find it hard to recognise close relatives from photographs?			
43	Does you child mistakenly identify strangers as people known to them?			
44	Does your child have difficulty understanding the look on your face?			
45	Does your child have difficulty naming common colours?			
46	Does your child have difficulty naming basic shapes such as squares, triangles and circles?			
47	Does your child find it hard to recognising well known objects such as the family car, classroom door?			
48	Does your child find it hard to find their way around a well known environment, for example school?			
49	Does your child find it hard to recognise an object if it is partially hidden or viewed from an unusual angle (such as a shoe under the bed with only the toe showing)?			
50	Does your child find it hard to spot which shoe is right and which shoe is left?			

51	Does your child find it hard to recognise people, or words, or objects if they are changed in any way, for example font of text, hairstyle?			
52	Does your child find it difficult to identify where sound is coming from, for example if they shout for you in the house do you have to clearly tell them which room you are in, as they are unable to work out where you are from your voice alone?			
	Thank you very much for your time.			

Appendix 12.2: English version of the Insight

Questions Inventoryand Tailored Visual Support

Strategies (VSS)

Q1 Trips / moves over toys and obstacles on the floor:

❶Keep floor space tidy and clear of obstacles. ❷Make a secret code to use when you areout and about. A tap on your child's shoulder could mean "Watch out! There is a hazard in the room." ❸Encourage your child to use a stick (maybe a hockey stick, or hiking pole or one picked up from the park) to feel the height of the ground ahead. The stick can also be used to identify and guide him around obstacles. ❹Try other ways to help your child walk over uneven ground and around obstacles. For example, offer your child your arm to hold.

Suggest he holds onto the belt or clothing of someone who is walking with him, or encouragehim to hold onto a wall. ❺Tell your child about obstacles and hazards he could miss due to his reduced field of view. (For example "there is a toy on the floor in front of you.") ❻whenyour child comes to an obstacle teach him to slow down, look, double check and then go - "Slow, Look, Check, listen, Go". This can be used inside and outside.

Q2 Has difficulty walking down stairs:

❶Use extra supports so your child can feel the height and position of steps. For example hand rails at the correct height (on both sides where possible) or holding onto an adult. ❷ Tell your child to slow down and hold on to the hand rails when he comes across steps. ❸ Give your child an agreed warning when

you come across steps or stairs, this could be a sound or touch. ❹Use bright lighting or spot lights on stairs. This is most important at the top, the bottom and on the landing. This will create shadows and make the steps clearer. ❺ Try using a torch at night. ❻Tell your child to look down at his feet when he goes down stairs. ❼Plain floor covering (e.g. laminate or plain carpet) can make it easier to walk downstairs. Patterns cause distractions, and can act like obstacles. ❽Try to keep walls plain. For example use plain not patterned wall paper and don't have too many pictures on the wall. ❾Let your child go first or last when using stairs. This is more important if there is a crowd of people (such as at school or in a shopping centre). ❿Encourage your child to use lifts or escalators. ⓫ Let your child have lots of time to practice stairs. ⓬ Arrange for your child toleave class a little earlier or later than his peers. This will allow him to miss the rush and crowds of other people on the stairs. ⓭ Talk to your child's school about having his classes on the ground floor. ⓮ Highlight the edge of each step with a bright colour. Or put a dot in the middle of each step to show where his foot should go. ⓯ Use a colour to mark the area beyond the top and bottom of the stairs. This may help to show more clearly where the stairs start and finish. ⓰ Give your child extra support when he is going up or down stairs/ steps.

Q3 Trips or are unaware of the edges of pavements going up/ down:
❶Give your child reminders. For example "There is a kerb to go up in 3 metres." ❷UsePedestrian Crossings because kerbs stones are lower at these points. ❸Give your child reminders to look down.

Q4 Appears to 'get stuck' at the top of a slide or hill:

❶Encourage your child to practice on small slides and/or by lying on his tummy on a scooter board or skate board. Some children choose to go down slides head first. Do not stopthis, but make sure it is safe. Children may do this because the upper part of their field of view is being used in this situation. ❷Give additional verbal information.

Q5 Looks down when crossing floor boundaries e.g. where lino meets carpet:

❶Give your child extra hints and prompts. For example "The carpet finishes here." ❷Trynot to use patterned carpets, plain carpets and floor surfaces may help make changes easier tosee. ❸Make sure floors are well lit where one surface stops and another starts. For examplewood to carpet at a doorway. ❹Try using coloured tape to mark a change in surface, this can make it easier for your child to spot the changes. For example, where carpet changes to lino. ❺Suggest your child pushes a tennis racquet, hockey stick, pram or other push toy ahead of him while walking.

Q6 Leaves food on their plate (near, far):

❶Put your child's favourite foods on the part of his plate that is usually forgotten. ❷At meal times make sure your child turns his plate. This could be done when you tell him, or when an egg timer has run out, or a stop watch reached a set time. ❸Put your child's plate on a spinning base, like those found in a Microwave. ❹Put food on plain, clear or transparent plates. ❺Try to avoid using gravy or sauces, as this can make the different foods on a plate look like one. ❻Sit your child on a lower seat at meal times. This may give

a better view of whole plate. ❼If you put more than one type of food on a plate make sureeach food is a different colour to the others. This may help your child to spot the different foods.

Q7 Leaves food on their plate (left, right):

❶Put your child's favourite foods on the part of his plate that is usually forgotten. ❷At meal times make sure your child turns his plate. This could be done when you tell him, or when an egg timer has run out, or a stop watch reached a set time. ❸Put your child's plate on a spinning base, like those found in a Microwave. ❹Put food on plain, clear or transparent plates. ❺Use different types and colours of food. This may make each food easier to spot. ❻Try to avoid using gravy or sauces, as this can make the different foods ona plate look like one. ❼Sit your child on a lower seat at meal times. This may give a betterview of whole plate. ❽If you put more than one type of food on a plate make sure each food is a different colour to the others. This may help your child to spot the different foods.

Q8 Has difficulty stepping into the bath, which is not related to balance:

❶Put one bathmat outside the bath and one inside. They should be bright, and a differentcolour to the bath. ❷Put stickers on the edge of the bath to make it stand out.

Q9 Has difficulty finding the beginning of a line or the next word when reading, or missespictures or words on one side of a page (left/right):

❶Ask your child to point to each word with his finger. ❷View only one

line of a book ata time. This can be done using a piece of card with a viewing window cut out of it. This reduces the amount of text on view and can be moved from line to line. ❸Make the font

size bigger and/or use double spacing between lines. ❹Make sure only a small amount of information is shown to your child at a time. For example, a small amount of text on a page, asmall amount of information on the board, enlarge the size of the print. ❺Make the print in text books bigger by using a photocopier to enlarge each page. ❻Block out extra bits of work or pictures on a page. This can be done using a blank piece of card or the back of your hand. ❼Use a magnifying glass or magnifying acetates. ❽Find the font size and number of words per page, and line, that your child prefers to work with (not the smallest he can read). Make sure work is given to your child in this size and layout. ❾Make a special work book by scanning photos, words and subjects that your child likes. ❿Mark the point where text starts so the eyes are drawn to it. This can be done with a ruler, the back of your hand or a brightly coloured dot. ⓫ Use a raised book stand for reading, for example a recipe book holder. ⓬ Practice reading using items that your child enjoys. For example, The children's newspaper "First news". ⓭ See if the class reading book is available as a large print book.

⓮ Break text into small bits. This can be done by enlarging text on a photocopier and thencutting it into small sections. ⓯ Use a computer programme which shows one word at a time.

Q10 Walks out in front of traffic:

❶Give your child extra hints. For example "We must wait here while we

check it is safe tocross." ❷When teaching your child to cross the road, teach him to cross at pedestrian crossings. ❸Remind your child to listen carefully for cars, as well as looking, when he is crossing the road. ❹Teach your child to turn his whole body from side to side when he looks for cars. You can encourage him by doing the same thing. ❺Give additional supervision and guidance at roads.

Q11 Bumps into doorframes or partly open doors (left/right/both):

❶Give extra hints. For example, "There is a door coming up in 3 metres." ❷Help door frames stand out by painting doors frames and skirtings a bright or contrasting colour from walls. ❸Replace doors with a beaded curtain. ❹If your child always bumps into the sameside of the door frame put a bright mark or picture on that side of door frame. Place the markor picture at your child's eye level.

Q12 Has difficulty seeing scenery from a moving vehicle:

❶Give extra reminders. For example "There is a church coming up on your left." ❷If your child doesn't like travelling in the car try asking him to wear wrap around sunglasses. This may reduce the feeling of movement and visual input. ❸When you are out and aboutvideo interesting things on a camcorder or mobile phone. These can be discussed with yourchild later. ❹Let your child sit in the front seat of the car.

Q13 Has difficulty seeing things which are moving quickly, such as small animals, children:

❶Tell your child things that he might want to know. For example, "Your friend Jenny is at the gate in a pink jacket." ❷Make sure friends and teachers

tell your child who they are, especially if they meet in a busy area with lots of movement. ❸Encourage your child to listen carefully to the voices of friends and family. Your child can then use a person's voice tohelp work out who they are. ❹Encourage your child to shout for the person he wants to find. ❺Encourage your child to use a mobile phone to call or text the person he wants to find. ❻If your child is planning to meet up with friends or family make sure a meeting point has been arranged before hand. ❼Teach your child to follow moving objects by moving his head as well as his eyes.

Q14 Has difficulty seeing moving water i.e. tends to over or under-fill a cup: ❶Ask your child to half fill cups. ❷Use a clear plastic cup. ❸Encourage your child to use a smaller cup that he can look into. ❹Practice pouring the liquid together. Count how many seconds of pouring it takes to half fill the cup. Then use this as a guide. ❺Try using a fluid indicator when pouring liquids. (This device hangs over the rim of a cup and makes a beep when the liquid reaches 4cm below the top of the cup). Fluid indicators are available from the RNIB. ❻When pouring cool liquids teach your child put the thumb of his non- dominant hand pointing down into cup. Your child will feel the liquid on his thumb and knowthat the cup is nearly full. ❼Teach your child to listen to the sound that is made when he fills a cup or glass. As a cup fills the sound of it filling changes. It becomes higher in pitch.

❽Mark the cup with nail polish at the level it

should be filled to.Q15 Has difficulty following a

cursor on the computer screen:

❶Make the cursor on the computer screen bigger. ❷Switch the cursor to a

flickering, instead of still, picture. This may make it easier for your child to find. ❸Vary size of the computer/TV screen to suit your child's ability to see the whole picture, a larger screen mayhelp if the child is needs a larger image, a smaller screen may help if the child finds it difficult to look at the whole picture at the one time.

Q16 Avoids watching fast moving TV, prefers to watch slow moving TV:

❶Try a flat screen TV as your child may find this easier to see.

❷Encourage your child tosit close to the TV. This may cut out some visual distractions. Some families have chosen to have an extra TV that their child can sit close to. ❸Reduce the number of objects around the TV. i.e. photos, pictures, patterned wall coverings. These all act as visual distractions.

❹Make sure the class teacher knows that many curriculum TV programmes can be difficultfor your child to follow. This is due to the speed and content of the programme. Your child

may be better off using the time for another task. ❺Old films (pre 1970 i.e. The Wizard of Oz) usually have simple graphics, fewer camera movements and less zoom photography. Andthey are usually slower moving. These factors may make older films easier for your child to follow. ❻Find programmes where the presenter sits or stands still (for example children's news, weather) and encourage your child to watch these.

Q17 Has difficulty catching a ball:

❶Practice catching skills with your child by throwing a balloon to each other. The balloonwill move slower than a ball and may be easier for your

child to catch. ❷Put a little bit of rice / water in the balloon. The balloon will make a noise as it moves so your child can hear where it is. ❸Use large, brightly coloured balls when playing catch or other ball games with your child. ❹Use balls with sound or light effects when playing catch or other ball games with your child.

Q18 When walking, holds onto your clothes, tugging down:

❶When walking together hold hands with your child. This works best if you hold your armstraight and slightly back. This gives your child a guide to the height of the ground ahead.

❷Talk to your child as he moves, tell him what is coming up. For example "In three steps you will be moving onto carpet, "You need to lift your feet higher here as the ground is bumpy." ❸Give your child extra physical support i.e. hand rails/banisters as required. ❹ Work on movement skills with activities and games. For example obstacle courses in the park, house or garden, riding, swimming or trampolining. ❺Try other ways to help your child walk over uneven ground and around obstacles. For example, offer your child your armto hold. Suggest he hold onto the belt or clothing of someone who is walking with him, or encourage him to hold onto a wall.

Q19 Finds uneven ground difficult to walk over:

❶When walking together hold hands with your child. This works best if you hold your armstraight and slightly back. This gives your child a guide to the height of the ground ahead.

❷Talk to your child as you walk together giving him reminders and

instructions. For example, "You need to lift your feet higher here as the ground is bumpy." ❸Encourage your child to use something to help with balance and tell him what is ahead. This could be a pram, wheeled toy, hockey stick or walking pole. Make sure the aid suits your child's age andis something he has chosen to use. ❹Make sure there is no change in height when one floor surface stops and the next begins. ❺Encourage your child to wear white trainers or shoes tomake his feet stand out. ❻In play grounds/ play areas use Astroturf or coloured matting.

Use different colours in different areas. ❼Use coloured rubber flag stones to help differentareas of ground in the playground or garden stand out more clearly. Make sure there is no height difference between each flag stone. ❽Make sure there is safe ground cover in outside areas that are frequently visited. For example, the playground. ❾Let your child practice balance skills by trying different activities, for example obstacle courses, wobble borads or horse riding. ❿Provide additional support and supervision if required.

Q20 Bumps into low furniture, such as a small table:

❶Remind your child to change his head position and look down if he wants to look at an obstacle. ❷Tell your child what is round about. For example "The table is just in front of you." ❸If you are moving furniture around make sure your child is involved. ❹Have lessfurniture in each room. This will make more space for your child to move around. ❺Make sure the furniture is a different colour to the floor. Use plain carpet and wall coverings. This will make the overall picture simpler for your child. ❻Make sure you do not

buy glassfurniture or furniture with sharp edges.

Q21 Gets angry if furniture is moved:

❶Remind your child to change his head position and look down if he wants to look at an obstacle. ❷Tell your child what is round about. For example "The coffee table is just in front of you." ❸If you are moving furniture around make sure your child is involved. ❹ Have less furniture in each room. This will make more space for your child to move around.

❺Make sure the furniture is a different colour to the floor. Use plain carpet and wallcoverings. This will make the overall picture simpler for your child.

Q22 Explores floor boundaries (e.g. lino/carpet) with their foot before crossing the boundaryor finds floor boundaries difficult to cross:

❶Tell your child about floor boundaries as he walks towards him. For example "The floorwill change to carpet in 3 steps." ❷Make sure there is good lighting in areas with floor boundaries (where one surface stops and the next starts). ❸Use plain floor coverings. ❹Give your child extra support if required. For example your arm, a rail, or the wall. ❺ Arrive early at new places. This may help your child get to know the new surroundings and find points where the floor surface changes.

Q23 Reaches incorrectly for objects, i.e. reaching beyond or around the object or whenpicking up an object, grasps it incorrectly, missing or knocking it over:

❶Use things that are a different colour to the surface of the unit. ❷Give extra reminders. For example, "The cup is just beside your elbow". ❸When picking something up use the same hand for guidance - touch the surface the object is on with the side of the hand and slide

the hand forward to pick up the object. Or use the other hand as a guide to find where the object is - touch the object with the non dominant hand allowing the reaching hand to use a more precise grip. The brain can note the position of the object through touch not vision. ❹Practice hand eye co-ordination with games and toys that develop this skill. For example Jenga, Snap, Wii, Eye toy for games console.

Q24 Has difficulty seeing something which is pointed out in the distance:

❶Try zooming in to an item of interest using a digital camera or camera phone. Consider recording the scene so you can talk about it later. Allow your child time to practice using a camera or phone to do this. ❷Give your child clear directions about where you would like him to look. For example "If you look at the big church, and then look past it you will see your friend." ❸Play "I spy" games. Encourage your child to chose a distant object and giveclues about it so that others have to find and identify the same object. (You could use a videocamera or digital camera to help your child find objects in the distance). ❹Encourage your child to look at the scenery regularly. Ask him to tell you what he sees and talk about it together. ❺Give your child lots of time to take in what he is seeing. Try not to rush him onto the next thing too quickly. ❻If you spot something your child would enjoy take him to the object so he can see it close up.

Q25 Has difficulty finding a close friend or relative who is standing in a group:

❶Let your child know who is around. For example, "your friend Jenny is at the gate in a pink jacket." ❷Make sure friends and teachers say who they are, especially if they meet your child in a busy place. ❸Point out friends and family in busy places such as the playground. ❹Encourage your child to listen carefully to voices to help locate friends andfamily. ❺Ask your child to call out for person he wants to find. ❻Ask your child to use mobile phone to call or text the person he wants to find. ❼Agree a meeting point before hand. ❽Use your voice to help your child find you. (For example, Mum calls out her child'sname so that her voice can be recognised as well as her face (be aware that some children find it hard to work out where a voice is coming from, so waving as well as shouting may help the child identify where Mum is). ❾Agree a meeting place in advance so your child knows where to find you. ❿Wear clothing that stands out and is easy to spot, for example apink jumper or luminous jacket. This item of clothing may then "pop out" at your child and help him to find you. Make sure tell your child about the item of clothing you plan to wear before hand so he knows what to look for. It is also important that the item of clothing can beseen from all angles. i.e. a jumper, not a badge.

Q26 Has difficulty playing team games with a lot of players, especially if there are a lot ofplayers and they are all moving:

❶Make sure that team members wear luminous/ brightly coloured bibs so that they can be seen more easily. The other team should wear different coloured bibs. ❷Ask team membersto keep shouting out where they are so

your child can spot them more easily. ❸Encourage your child to try sports that involve fewer people and less movement. For example golf or fishing.

Q27 Has difficulty finding an item in a supermarket e.g. finding the cereal or magazine:

❶Give extra spoken help and clear instructions. For example "It is in the middle shelf at thefront." ❷When you visit shops ask your child to find a few things. To start with always ask him to find the same things. Then slowly add in extra items. This may help your child remember where things are and learn how to find his way around the shop. ❸Make sure your child does not have to look through too many items to find what he is looking for. This can be done by shopping in small shops with small numbers of items on each shelf. ❹Put only one type of clothing in each drawer or cupboard section. Make sure drawers are not toofull. For example, have one drawer for socks, one for t-shirts etc.

Q28 Has difficulty locating an item of clothing in a pile of clothes:

❶Put only one type of clothing in each drawer or cupboard section. Make sure drawers arenot too full. For example, have one drawer for socks, one for t-shirts etc. ❷Try different storage styles and layouts. For example, order clothes across the way or down the way - underwear on the top or far left box, t-shirts in the next box, trousers or skirts in the next andso on. ❸Hang clothes on a rail or in a wardrobe rather than fold them. ❹Try hanging clothes in groups, put items of the same type and colour together. i.e. Red tee shirts together,blue tee shirts together, black trousers together. ❺Only put one type of clothing in each drawer. Don't overfill drawers. i.e. have one

drawer for socks, one for t-shirts etc. ❻Hang all the clothes for one outfit together. ❼Lay out clothes the night before. ❽Make sure thewalls, carpets and bed covers are plain. This means there is less for your child to 'take in' when he is looking for something. ❾Use spotlights or a lamp to light up important areas inroom i.e. above drawers. ❿Involve your child in sorting out his belongings, let him decide where things should go.

Q29 Has difficulty selecting a chosen toy in a full toy box:

❶Store shoes in an elevated shoe rack. ❷Involve your child in sorting out his belongings.Try not to use too many storage systems. ❸Put just a few items in each toy box, bag, drawer, cupboard and on each shelf. ❹Encourage your child to put things back in the same place when he has finished using him. ❺Label storage boxes and drawers with photos or pictures of what is inside. Try colour coded boxes, for example console games in blue boxes, trains in red boxes, dolls in yellow boxes etc. ❻At school make sure there are only a few things on your child's desk, in his pencil case and in his bag. ❼Try using 'see through' containers. i.e. pencil case and school bag. ❽Use clearly labelled storage so that objects canbe easily put away after use. This will reduce the number of things your child is trying to see at the same time (visual clutter). ❾Set up a special area in your child's room for him to put important items i.e. a box or plate on the bedside table for glasses, I pod, mobile phone, TV remote. Encourage your child to always put things back in this place after he has used him.

❿Have colour coded books/ folders for each subject.

Q30 Has difficulty identifying objects when they are on a similar background such as a whitetee shirt on a white sheet:

❶Use contrasting, plain colours and backgrounds.

Q31 Gets lost in places where there is a lot to see, e.g. in a crowded shopping centre:

❶Make sure you have agreed a meeting point with your child, in case he gets lost while outand about. ❷Ask your child to stay close to the adult looking after him when they are out and about. ❸Practice reading and using simple maps with your child. ❹Give extra practice to help your child find his way around new and unfamiliar places. This may help your child get to know the area. ❺Help your child to find his way around a new place. Youcan do this by encouraging your child to go a short distance and then return to you, graduallyincrease the distance travelled and the number of left / right turns. This could be used at a party venue or supermarket. ❻Use walkie talkies so that your child can keep in touch, but can also develop his independence.

Q32 Finds copying words or drawings time-consuming and difficult:
❶Try Voice Activated soft-ware, which will read text and emails to your child. ❷Reduce the amount of copying your child has to do by putting information on a pre-printed sheet. ❸Email or scan information to a laptop instead of asking your child to copy it. ❹Make sure words are clearly visible to your child. The size of print needed will depend on how clearly your child sees (visual acuity) and how well he sees contrast (contrast sensitivity). ❺Find out what colour of ink and size of print is clearest for your child to see. You should do this for both computer and board work. ❻Reduce the

amount of information on the board.

Leave only the most important text. ❼Make sure the space around the board is clear from anything that could distract from the board itself. i.e. pictures, mobiles. ❽In class make sure that your child sits face on to the board. Your child should sit at the distance he finds it easiest to read the board. ❾Make sure the board is well lit. ❿Let your child to use a mobile phone to photograph writing from the board. This will save him from having to writeit down. For example, homework instructions. ⓫ Play games that use your child's ability toremember what he has seen. For example, 'I spy', 'tell me what you saw'. ⓬ Make use of your child's memory for things he hears. For example, speak instructions out loud as well as writing them down. ⓭ Do not ask your child to do more than one thing at a time, for example write and speak or write and listen at the same time. ⓮ Scribe work for your child where possible. This can need a lot of practice, especially if you are scribing for maths, science or languages.

Q33 Has difficulty reading crowded text on paper or on a computer screen, but can copebetter if some of the text is covered or removed:

❶Hide text around the words your child is trying to read using a typoscope (a piece of black card with a viewing slot in it). This can be moved along each line and from line to line.

❷Make words (font size) bigger. This will reduce the number of words on the paper or screen at any one time and may make it easier for your child to 'take in' the information. ❸ Make sure your child is not asked to 'take in' too much information at the same time. This canbe done by putting only a few things on

each page or on the classroom board. ❹You can trydifferent line spacing, font style and colour contrast. You should be aiming to find the layout that allows your child to read the fastest. Once this layout have been found use it as often as you can. Note you may need books to be scanned and enlarged or converted to double spacing. ❺Make sure the computer set up is the same every time it is turned on.

Q34 Has difficulty finding letters on keyboard but knows alphabet:

❶Make key-board keys clearer using brightly coloured alphabet stickers. ❷Use a larger key-board with bigger keys. ❸Try using an adapted mouse or keyboard. For example, keyboard with a finger guard or highlighted keys, or a mouse that is enlarged. ❹Let your child try different computer programmes to reduce the dexterity skills required i.e. clicker 5 clicks on the word, your child will then have to find the word and click with the mouse, not type each individual letter with his fingers. ❺Try Voice Activated soft-ware. The key-boardwill then play a less important role in computer work.

Q35 Sits closer to the television than about 30 cm:

❶See if your child finds it easier to watch TV on a large screen TV. ❷Make sure there are no objects (visual clutter) between your child's seat and the TV. ❸Do not put things around the TV. These objects (photos, plants etc) can create 'visual clutter' and increase the amount of information your child is trying to 'take in' at once. ❹See if your child finds a smaller TV easier to watch. A large screen can make it hard to follow what is happening in all parts of the picture. ❺Join a film or book club to get older,

slower moving films. Theseusually have fewer special effects.

Q36 Finds it difficult to keep to task for more than 5 minutes, or after being distracted finds itdifficult to get back to what they were doing:

❶Take away unnecessary objects that clutter your child's work area. ❷If possible, take other distractions away from your child's work area. i.e. sound, movement. ❸Ask your child to work in short bursts. You can do this by giving tasks in short blocks and breaking down activities. ❹Time your child's concentration span. Work with your child only for as long as he can concentrate. You may be able to build this up slowly. ❺Use a timer to set time limits for tasks. This should be based on your child's concentration span. The timer can be one that you watch (eg an egg timer) or one that makes a noise (eg a buzzer). ❻Vary tasks and demands. i.e. seating, standing, moving, listening, talking, looking. ❼Give your child lots of reminders and praise. For example "You are doing well. Just 1 more minute to go". ❽With your child, make up an unspoken signal that you can give to help your child get back on task. For example, a tap on the shoulder or two claps. ❾Have a quiet work space for your child at home and in class. Don't have too many things in this space. Too many objects can act as 'visual distractions' and make it harder for your child to find what he is looking for. This work space should not be beside a window. ❿Let your child have lots of breaks. Let him move around at these times. For instance he could hand out pencils or go for a message. your child may need to do something active before he gets started on a writing task. ⓫ Make sure your child sits near the front of the class, facing the board and teacher.

But make sure your child does not sit at the very front of the class, as he may want to turn around. Sitting near the front of the class may take away 'visual distractions' and the need tolook over heads. ⓬ Let your child try using a chair with arms. This may help him to balance. ⓭ Let your child use a fidget object to help him focus on what is being said. This could be "blu tac", a giant paper clip, or a rubber. ⓮ Let your child use head phones or earplugs so noise does not disturb him.

Q37 Reacts angrily when other restless children cause distraction:

❶If possible, take other distractions away from your child's work area. i.e. sound, movement. ❷Let your child use head phones or ear plugs so noise does not disturb him.

❸See how your child gets on sitting at a separate desk at the end of the group. This may give him more space without leaving him out of the group. If your child has visual attention problems on one side, consider sitting with the affected side next to the wall (i.e. if your childalways leaves the food on the right side of his plate or bumps into the right side of a door frame sit him with his right side to the wall).

Q38 Bumps into things when walking and having a conversation:

❶Let your child know what is coming up. For example "There is a tree coming up in front of you." ❷Ask your child to carry out one task at a time. For example, walking or talking orlistening.

Q39 Misses objects which are obvious to you because they are

different from theirbackground and seem to 'pop out' e.g. a bright ball in the grass:

❶Do not present too many items at one time. ❷Space the items out. ❸Make objectsstand out from their background. This can be done by having plain floors, walls and bed spread. ❹Make sure objects are brightly coloured and a different colour to their background. This can make them 'pop out'.

Q40 Becomes distressed in places with a lot of clutter or busy environments, such assupermarket or shopping centre:

❶Do not have too many bits and pieces in areas that your child uses a lot. For example, photos on the wall or mobiles in class. ❷Be prepared for difficult behaviour in busy placesand take steps to make the situation easier. For example, give lots of advance and warnings, only go to these places for a short time have a reward for afterwards. ❸Help your child to use his other senses. For example, let him listen to an I pod or MP3 player. ❹Give your child something to feel. This will use his sense of touch. he could use a fidget object, toy, rubber or giant paper clip. ❺Give your child something to suck or chew. This will use his sense of taste. You could use chewing gum, mint, lemon or an ice lolly. ❻Give your child something to sniff. This will use his sense of smell. You could use a hanky with some perfume on it. ❼Ask your child to push the trolley or carry the basket. These activities use deep muscles and can be calming. ❽Ask your child to help you. For example, when shopping ask him to find things, make sure he has his own job at assembly. ❾Let your child to take lots of breaks to move around. ❿Make sure there is

a quiet area for your childto use, both at home and at school. This should be a space with very few objects in it. ⓫ Practice in smaller, quieter areas. For example a small dining room, or a small shop rather than the large school dining room or large shopping centre. ⓬ Gradually build up your child's experience of new places. For example, start by taking a trip to a small shop for one item. ⓭ Go to parties early. This will give your child time to get to know the new surroundings.

Q41 Has difficulty recognising close relatives in real life:

❶Wear clothes that make you easy to spot. For example, a bright pink top or scarf. The item of clothing must be seen from all angles. Tell your child the item of clothing you will be

wearing so he knows what to look for. ❷Teach your child to look for special marks on people that he knows. For example, birthmarks or tattoos. ❸Practice recognising different voices with your child. ❹Let your child know that you are there. For example, shout your child's name. ❺Make sure people know your child finds it hard to recognise faces. Ask people to say hello, and who they are, when they meet your child. ❻Make sure your child has a buddy or friend to help him find people. ❼Ask your child to shout out the name of the person he is looking for.

Q42 Has difficulty recognising close relatives from photographs:

❶Use photos of close family and friends to practice spotting people. ❷Use pictures withonly one person to practice identifying people. ❸Give your child clues. For example, "Wesaw them here yesterday."

Q43 May mistakenly identify strangers as people known to them:

❶Give your child extra clues. For example, "It can't be your Gran because she is at home."

❷Remind your child to look again.

Q44 Has difficulty understanding the meaning of facial expressions:

❶Make sure people spending time with your child know it is difficult for him to recognise people. Give extra help and hints. ❷Find out if your child can tell when you are happy/ sad/cross etc by the look on your face (facial expression). If he can recognise facial expressions try and work out the easiest distance from him to do this. ❸Ask your child to listen carefully to the tone of voice, and words being used. This may help him to work out the mood of the person he is talking to. ❹Practice recognising different facial expressions with your child. i.e. Happy, sad, cross, surprised. ❺Use words and tone of voice that match the expression on your face. For example "I'm happy, I'm smiling at you". Ask every one

spending time with your child to do this. ❻Exaggerate your voice and use expressed emotions to help your child to understand. For example, by acting very happy, very sad, veryangry.

Q45 Has difficulty naming common colours:

❶Find out if this is due to a problem with colour vision (colour blindness).

❷Practice the names of the primary colours (red, blue and yellow) using objects that your child knows well.

Q46 Has difficulty naming basic shapes such as squares, triangles and circles:

❶When learning shape names use 3D models. ❷Ask your child to touch the shape and say the name. ❸Practice using 3D shapes. ❹Ask your child to feel objects with his hands.

❺Ask your child to make shapes with his fingers in sand or shaving foam.

❻Make sureyour child does not have to look for too many things at the same time. ❼Encourage your child to play computer games that use recognition skills.

Q47 Has difficulty recognising familiar objects such as the family car, classroom door:

❶When your child has to find the item that belongs to him from a group of similar items, mark the one that belongs to him with something easy to spot. For example, put a plant on thedoorstep of your house so your child knows which house is home, put a hanging toy on the rear windscreen of the car or a cushion on parcle shelf so your child can identify the family car, make sure there is a poster or special mark on his classroom door. ❷Agree a meeting point in case your child gets lost. ❸Ask your child to stay close to the adult he is with. ❹ Help your child learn how to find his way around. This can be done by going a little way and then going back to where you started. Gradually build up distance and the number of left / right turns. ❺Try using a walkie talkie. This will let you stay in touch while letting you child get used to finding his way without help.

Q48 Has difficulty navigating familiar environments, e.g. school:

❶Use brightly coloured tape to mark your child's desk, this may make it easier to find. ❷If your child gets lost in places he knows (for example, home or school) use a few circle markers, or brightly coloured footprints on the

floor, to mark important routes i.e. seat to board, seat to door, hall to bedroom.

Q49 Has difficulty recognising an object if it is partially hidden and not fully visible, orviewed from an unusual angle:

❶Make sure all items are easy to see and do not over lap. ❷Store items upright. ❸Try araised shoe rack. ❹Make an organised work space for your child. Mark lines on the desk with coloured tape for a book, pencil etc. Don't keep too many things on the desk. ❺Make sure your child has a schoolbag with a pocket for each item/ group of items. ❻Try giving your child a flat pencil case with a clearly marked space for each item.

Q50 Has difficulty recognising right and left shoes:

❶Mark the shoe of your child's dominant side (right if he is right handed, left if he is left handed) with a sticker, or decorative bead or initials. This will show which one should go onfirst.

Q51 Has difficulty recognising people, word, objects if changed in presentation style, i.e. fontof text, hairstyle:

❶Try to approach your child face on. ❷Make sure all text given to your child is set out inthe same way. i.e The same font, line spacing and colour. ❸Tell your child if you change the way you look. For example, a new hairstyle.

Q52 Finds it difficult to identify where sound is coming from .i.e. if they shout for you in thehouse do you have to explain which room you are in:

❶If your child calls for you always say what room you are in. ❷Use actions to show where you are. For example, wave. ❸Ask your child to use a mobile phone to call or textyou when he is

looking for you.

Appendix 12.3: Modified Insight Questions Inventory and visualsupport strategies (Nigerian version)

About your child

Child's forename:	Surname:
D.O.B.:	Gender:
Instructions to parents	
These questions are designed for a range of ages, so somequestions may seem odd Your child may have difficultywith some behaviours listed below but not others – this is normal.	
For each of the items listed, please could you click on the box next to it and select which best fits with your child's present behaviour: never/ rarely/ sometimes/ often/ always/ not applicable (NA).	

1 Does your child fall or stumble onto things on thefloor?

2 Does your child findit hard to walk downthe hill or stairs?

3 Does your child want to fall or does it seem as if your child does not notice when the foot path or pavements go up ordown?

4 Does your child get frightened not knowing whatto do at the top of a slide or hill or in front of a gutter?

5 Does your child look down when they cross floorboundaries, for example where tiles meets the cement floor?

6 Does your child leave food on their plate? If so,is this on the near or far side?

7 Does your child leave food on their plate? If so,is this on the left or right side?

8 Does your child have any problem stepping intothe bath, apart from any problems with being able to stand by himself well-balance?

9 Does your child find it hard/difficult to find the beginning of a line or the next word when reading, or miss pictures or words on one side ofa page (left /right)?

10 Does your child walk, move out in front of traffic(coming from left/ right / both)?

11 Does your child hit his/her body into door framesor partly open doors (left/right/both) when tryingto pass through?

12 Does your child have difficulty/hard seeingthings from a moving vehicle?

13 Does your child find it hard to identify things (orknow what has just passed by) that are moving pass quickly, such as children, small animals?

14 Does your child find it hard to see moving waterin a container i.e. to know when the cup is full?

15 Does your child find it hard to follow the arrowhead on the computer screen?

16 Does your child avoid watching fast movingvideo CD, and prefer to watch slow moving video CD programmes?

17 Does your child find it hard to catch a ball?

18 When walking, does your child hold onto yourclothes, dragging down?

19 Does your child find uneven ground hard to walkover?

20 Does your child run into low furniture, forexample a small stool?

21 Does your child get angry if furniture is moved?

22 Does your child explore floor boundaries (for example, where tiles meets a cement floor) withtheir foot before crossing

23 Does your child reach incorrectly for objects (i.e.reaches beyond or around the object or when picking it up, grasp incorrectly, missing or knocking the object over)?

24 Does your child find it hard to see somethingpointed out in the distance?

25 Does your child find it hard to know a relative orfriend who is standing in a group?

26 Does your child find it confusing or difficult when playing team games with a lot of players,especially if there are a lot of players and everyone is moving?

27 Does your child have difficulty finding an item inthe kitchen or cupboard shelf e.g. finding the salt or pepper?

28 Does your child find it hard to find an item ofclothing in a pile of clothes?

29 Does your child find it hard to find something they want in a full toy cartoon, shoe in a box ofshoes etc?

30 Does your child find it hard to find anything when the object is in a place with the same color of the place? E.g. awhite tee shirt on a white bedsheet?

31 Does your child get lost in places where there is alot to see, e.g. in a crowded market or church?

32 Does your child findcopying words or drawings time consuming and difficult?

33 Does your child have difficulty reading crowded text on paper or on a computer screen, but can cope better if some of thetext is covered or taken away?

34 Does your child find it hard to notice or look forletters on a keyboard but knows the alphabet?

35 Does your child sit closer to the television thanabout 30 cm?

36 Does your child find it difficult to concentrate orfocus or perform a task for more than 5 minutes,

or after being distracted do they find it difficult toget back to what they were doing?

37 Does your child reactangrily when other children cause distractions or making noise or movements all around?

38 Does your child hit into things when they arewalking and talking at the same time?

39 Does your child miss objects that are obvious to you because they are differentfrom their background and seem to ‘jumpout’ e.g. a bright ball in the grass?

40 Does your child get angry in busy places,such as markets or church?

41 Does your child findit difficult to recognise close relatives in real life?

42 Does your child findit hard to know who their aunties and uncles’ or sisters andbrothers are from photographs?

43 Does your child mistakenly identifystrangers as peopleknown to them?

44 Does your child havedifficulty understanding the meaning on your face?

45 Does your child find it hard to name commoncolours?

46 Does your child have difficulty naming basicshapes(call the names of the shapes) such as squares, triangles and circles?

47 Does your child find it hard to recognise well known objects such as the family house or the church building?

48 Does your child find it hard to find their way around a well known environment, for exampletheir home or church?

49 Does your child find it hard to recognise an object if it is partially hidden or viewed from an unusual angle (such as a shoe under the bed withonly the toe showing)?

50 Does your child find it hard to identify whichshoe is right and which shoe is left?

51 Does your child find it hard to recognise people, or words, or objects if they are changed in any way from what they are used to, for example fontof text, hairstyle?

52 Does your child find it difficult to identify where sound is coming from, for example if they shout for you in the house do you have to clearly tell them which room you are in, as they are unable to work out where you are from yourvoice alone?

Appendix 12.5: Example of an Insight question inventory question and corresponding visual

support strategy

Insight question 10: Does your child walk or move out in front of traffic(coming from left/ right / both) or someone coming in front of him?

Insight visual support strategy (IVSS)

❶ Give your child extra hints. For example, "We must wait here while we check it is safe to cross." ❷ When teaching your child to cross the road, teachhim to cross at pedestrian crossings or sidewalks. ❸ Remind your child to listen carefully for cars, as well as looking, when he is crossing the road. ❹ Teach your child to turn his whole body from side to side when he looks for cars. You can encourage him by doing the same thing. ❺ Give additional supervision and guidance at roads.

IQI No 7: Does your child leave food on their plate? If so, is this on the left or right side?IVSS No 7

❶Put your child's favourite foods on the part of his plate that is usually forgotten. ❷At meal times make sure your child turns his plate and you show him how to do so too. This could be done when you tell him, or when an egg timer has run out, or a stop watch reached aset time. ❸Put your child's plate on a spinning base, like those found in a Microwave. ❹ Put food on plain, clear or transparent plates. ❺Use different types and colours of food.

This may make each food easier to spot. ❻Try to avoid using gravy or sauces, as this can make the different foods on a plate look like one. ❼Sit

your child on a lower seat at meal times. This may give a better view of whole plate. ❽If you put more than one type of food on a plate make sure each food is a different colour to the others. This may help your child tospot the different foods.

12. 6 Questionnaire for the systemic and ocular examination of CP

12.6.1 CEREBRAL PALSY EXAMINATION QUESTIONNAIRE

Date form updated:						

Name of KI	

Phone number of KI	

Local government Area	1,2,3,4,5,6,7,8,9,10,11,12,13, 14,15,16, 17,18,								
Date of assessment		D	D	M	M	Y	Y	Y	Y

Site of assessment (Primary Health Centre)		
Source of patient	key Informant = 1 Hospital records = 2 School = 3 Other = 4	

Demographic details:

First name			
Family name			
Date of birth		D D M M Y Y Y Y	
Age (years) at last birth day			
Sex	Male=1 Female=2		
History taken from?	Mother=1 Father=2 Grandparent=3 Sibling=4 Child=5 Other=6		

Village name			
Mobile phone number			
Tribe	Breakdone to the 5		

Religion	Christian=1 Muslim=2 Spiritualist=3 Traditionalist=4 Other (state)=5		
Mother's Marital status as of today?	Married=1 Widowed=2 Divorced=3 Single (never married)=4 Separated=5 Not known=6 Child orphaned/left with family=7		

Who is the head of the family?	Father=1 Mother=2 Grandparents=3 Uncle=4 Aunty=5 Other=6		
Resident in LGA for last 12 months?	No=0 Yes=1		

History

Clinical history:

Does your child have any problems? What about eye problems?What are your child's problems?(As described by the career)

1.

2.

3.

4.

When did they start?

How did it start?

Is the child slow to learn? 1. Yes 2. No

Does the child have problems with behaviour? 1. Yes 2. No:

Does your child convulse? 1. Yes 2. No

Treatment History:

Have you ever been given a diagnosis for the problems?	No=0 Yes=1 NK=99	
Did you go to get treatment for the problem?	No=0 Yes=1 NK=99	What exact problem is been referred to?
If no, why not?		
Why?		
Details:		

Do you go for regular check ups?	No=0 Yes=1 NK=99	
Are they taking any medication?	No=0 Yes=1 NK=99	
Current treatment?		Dose/Timing
How long have they been taking any drugs (months)?		

Previous treatment:

Has the child been immunized?	No=0 Yes=1 NK=99

if Yes, for?	OPV1 OPV2 OPV3 VIT A 1 VIT A 2 DPT1 DPT2 DPT3 Measeles HIN Others All
Do they continue to have traditional treatment now?	

Past medical history:

Was mother healthy in pregnancy?	No=0 Yes=1 NK=99
Details:	
Was mother exposed to the following during pregnancy?	No=0 illicit drugs =1 toxins=2 infections=3 maternal diabetes=4 acute maternal illness=5 trauma=6 radiation exposure=7 Herbal remedies= 8 NK=99
Was the labour at the right time and normal?	Premature=1 Prolonged=2 Normal=4 NK=99

Details	
Where was the child born?	Hospital=1 Dispensary/Health centre=2 Home=3 TBA=4 Church=5 Other=4 NK=99
Details:	
Was the delivery normal?	SVD=1 CS=2 Forceps=3 Vacuum=4 Hand assisted=5 NK=99
Details:	
Did the baby cry immediately?	No=0 Yes=1 NK=99
Was the baby blue at birth?	No=0 Yes=1 NK=99

Did the baby turn yellow?	No=0 Yes=1 NK=99
Did the baby suck well after birth?	No=0 Yes=1 NK=99
Was the baby admitted after birth?	No=0 Yes=1 NK=99
Any fever in first few weeks?	No=0 Yes=1 NK=99
Any seizures as a baby?	No=0 Yes=1 NK=99
Were there adverse perinatal events? Did anything happen around the time the child was been born?	No=0 Yes=1 NK=99
Any abnormal softness of the body when baby was born	No=0 Yes=1 NK=99
When was this noticed?	

Any abnormal stiffness of the body	No=0 Yes=1 NK=99
When was this noticed?	
When did they say their first words	2yrs=1 >2yrs=2 Never=0
Do they startle to loud noise :	Yes =1 No=2 NK(Not known) =99
Is there a problem with the childs hearing:	Yes=1 No=2 not known=99
Did the child crawl	Yes=1 No=2 Nk=99
Head control at 4months	No=0 Yes=1 NK=99
Rolling at 6 months	no=0 Yes=1 NK=99

Sitting 10 months	no=0 Yes=1 NK=99
Walking at age 18 months	no=0 Yes=1 NK=99
Did the child have any serious illnesses in the past?	No:0 Yes:1 NK=99
Seizures?	No=0 Yes=1 NK=99
Malaria with seizures?	No=0 Yes=1 NK=99
Malaria with coma?	No=0 Yes=1 NK=99
Meningitis/encephalitits?	No=0 Yes=1 NK=99
Tuberculosis?	No=0 Yes=1 NK=99

Admitted with pneumonia?	No=0 Yes=1 NK=99
Sickle cell disease?	No=0 Yes=1 NK=99
HIV o	
Details of FHx of CP:	
Is there a family history of this problem?	No=0 Yes=1 NK=99
In brother?	No=0 Yes=1 NK=99
In sister?	No=0 Yes=1 NK=99
In father?	No=0 Yes=1 NK=99
In mother?	No=0 Yes=1 NK=99

In grandparent?	No=0 Yes=1 NK=99
Are parents consanguineous?	No=0 Yes=1 NK=99
Details family history	

Childs equipment use:

Does the child move about in his environment?	No=0 Yes=1 NK=99	if yes how?.....
hearing aid	No=0 Yes=1 NK=99	
wheel chair	no=0 Yes=1 NK=99	
walker	no=0 Yes=1 NK=99	

stick	no=0 Yes=1 NK=99	
crutches	no=0 Yes=1 NK=99	
Standing frame	no=0 Yes=1 NK=99	

Schooling:

Has the child ever gone to school?	No=0 Yes=1 NK=99	What school?
What school is this child in?		
What class is the child currently in?		
Age (yrs) they started school	NK=99	
Highest level achieved?		
Are there any visual problems at school?		
What problems?		

Are there any behaviour problems at school?	No=0 Yes=1 NK=99	
What problems?		
Is he/she performing well at school?	No=0 Yes=1 NK=99	
When last (months, years)did they go to school?	…	Why did they drop?
What position is he/she in the class? (NK=99)		/
Did they repeat a year?	No=0 Yes, only once=1 Yes,more than once=2 NK=99	
Why?		
Is educational level appropriate for age?	No=0 Yes=1 NK=99	

Do they attend school regularly now?	No=0 Yes=1 NK=99	
If no, why?	Epilepsy=1 Financial=2 Social=3 Other=4 Physical disability=5 Other ill health problems=6 (name .....) NA=99	
Is educational level appropriate for age?	1. yes 2. No 99. NK	
Do they attend school regularly now?	. Yes 2. No 99. NK	
If no why?	1. epilepsy 2. Financial 3. Social 4. Other problems 5. Physical disability 99. Not applicable	

Knowledge and attitudes: Person answering:

Why do you think the child behaves the way they do?	Hereditary/genetic =1 Sprirtual reason=2 Natural forces=3 Mental illness=4 Food/drink=5 Lunar phases=6 Physical illness=7 Alcohol=8 Problem in the brain =9 Infections =10 Hunger/poverty=11 Emotions=12 Don't know=13 Other = 14 NK=99	
If spiritual reason, which?	Will of god/allah =1 Witchcraft /sorcer=2 Punishment =3 Ancestors =4 Possession by spirits=5 Other=6 NA=99	

Does career think visual problems will get worse?	better=0 worse=1 Stable=2 NK=99	
Why?		
What should be done abut the eye problems noticed?		
Why?		
Any activities avoided/limited because of vision problems?	No=0 Yes=1 NK=99	
What activities?		
Why avoided?		
Has relationship with others been affected?	No=0 Yes=1 NK=99	

With whom?	other family members =1 friends=2 carer's spouse/partner=3 others in village/community=4 childs teacher =5 NA=99	
Why?		
Are you aware of any group of people that can help you with this problem?	No=0 Yes=1 NK=99	If yes, please name the group:.....

Examination

EXAMINATION		
Weight (kg)	NK=99	
Height (cm)	NK=999	
OFC (CM)	NK=99	
MidArmCircufrence (MCM)	NK=99	
Handedness	Left=1 Right=2 NK=99	

Hearing	Normal=0 Abnormal=1 NK=99	
Dysmorphism (abnormal body efeatures)	No=0 Yes=1 NK=99	
Details:		
Blood pressure (mmHg)	Systolic/diastolic (NK=999/999)	/
BP abnormal?	No=0 Yes=1 NK=99	
Respiratory system	Normal=0 Abnormal=1 NK=99	
CVS system	Normal=0 Abnormal=1 NK=99	
Abdominal examination	Normal=0 Abnormal=1 NK=99	

Any burn marks on the childs body?	No=0 Yes=1 NK=99	
Any marks on the childs body	No=0 Yes=1 NK=99	
Details of systemic examination:		
Memory®no. out of 5) objects remembered after 15 seconds		
Tongue deviates?	No=0 Yes=1 NK=99	
Arms wasted?	No=0 Yes=1 NK=99	
If arms wasted	Right=1 Left=2 Both=3 N/A=99	

Tone in arms	Normal=0 Increased right=1 Increased left=2 Increased both=3 Decreased both=4 NK=99	
Contractures in arms	No=0 Yes=1 NK=99	
Power in arms	Normal=0 Abnormal=1 NK=99	
If abnormal	Decreased on right=1 Decreased on left=2 Decreased proximally(L+R)=3 Decreased distally(L+R)=4 Decreased both =5 NA=99	
Legs wasted?	No=0 Yes=1 NK=99	
If legs wasted	Right=1 Left=2 Both=3 N/A=99	

Tone in legs	Normal=0 Increased right=1 Increased left=2 Increased both=3 Decreased both=4 NK=99	
Contractures in legs?	No=0 Yes=1 NK=99	
Power in legs	Normal=0 Abnormal=1 NK=99	
If abnormal power	Decreased on right=1 Decreased on left=2 Decreased proximally(L+R)=3 Decreased distally(L+R)=4 Decreased both=5 NA=99	
Reflexes		
(N=normal, I=inc, D=dec,NK) Biceps Knee Ankle Plantar	R	L

If abnormal	Increased in legs>arms =1 Increased in all limbs=2 Increased on right=3 Increased on left=4 Decreased in all=5 Normal/NA/NK/OTHER=99	
Co-ordination	Normal=0 Abnormal=1 NK=99	
Gait	Normal=0 Abnormal=1 NK=99	
Details:		
Systemic examination	Normal=0 Abnormal=1 NK=99	
Neurological examination	Normal=0 Abnormal=1 NK=99	
Neck control	No=0 Yes=1 NK=99	

Trunk control	normal=0 Abnormal=1 NK=99	
Type of CP	1. Spastic 2. Spastic bilateral 3. Spastic unilateral 4. Dyskinetic 5. Choeoathetois 6. Diplegic 7. Unclassified 8. Ataxic	
Anatomic distribution		
CP side	1.left 2.right 3. both	
Timing of event causing CP		
Suitable for CT scan	No=0 Yes=1 NK=99	

Classification of GMFS type:	Monoplegia (5%– - one limb, v rare=1 Hemiplegia (41.5%)- one side=2 Diplegia (25%– - lower limbs (often =3asymmetric) UL involvement minimal Triplegia (7.5%– - three limbs, v rare=4 Quadraplegia (21%)- all limbs, most =5 severe involvement Unclassifiable=11 Other =12 NK=99	
GMFSC	Grade1=1 Grade2=2 Grade3=3 Grade4/5=4	
malnutrition	No=0 Yes=1 NK=99	
Swallowing difficulty	normal=0 Abnormal=1 NK=99	

Can they feed themselves	normal=0 Abnormal=1 NK=99	
Do they cough/choke on food or fluid	yes=1 no=2 nk=0	
Do they take more tha½/2 hr to feed	No=0 Yes=1 NK=99	
Do they eat with other children at the same time from the same bowl?	No=0 Yes=1 NK=99	
Likely aetiology	Prenatal=1 Perinatal=2 postnatal=3 Undetermined=0	
Co-morbidity	No=0 Yes=1 NK=99	
Feeding difficulties	No=0 Yes=1 NK=99	

Swallowing difficulties	No=0 Yes=1 NK=99	
Seizure disorder	No=0 Yes=1 NK=99	
Inattention (ADD)	No=0 Yes-1 NK=99	
Drooling	No=0 Yes-1 NK=99	
Intellectual impairments	No=0 Yes-1 NK=99	
Cognitive impairments	No=0 Yes=1 NK=99	
Hearing impairments	No=0 Yes=1 NK=99	
Disorders of speech	No=0 Yes=1 NK=99	

Emotional and personality derangement	No=0 Yes=1 NK=99	
Learning disability	No=0 Yes=1 NK=99	
Walking	No=0 Yes=1 NK=99	
Type of co-morbidity	Behavioural problems=1 Motor difficulties=2 Visual impairment=3 Hearing impairment=4 Cognitive impairment=5 Other=6 Write in words……. More than 1=7 Feeding difficulties=8 Seizure disorder: 9 NA/NK=99	
FINAL DIAGNOSIS		

Field Plan

Plan for management/follow-up Refer to: 1. Study Trial 2. FGD 3. In depth 4. Investigation 5. Refer to UCTH pediatric ward 6. Refer to UCTH childrens eye clinic 7. Refer for counselling 8. Refer to physiotherapist 9. Refer to optician on field		
CT/MRI head done	No=0 Yes=1 NA=99	Date:

Name of Physician:

Signature of Physician:

12.6.2 Physiotherapy Assessment form

Name: Sex:

Child ID Code: Date of Birth:

Address:

Phone:

1. Diagnosis by the doctor: CP/ CP with

Assessed by: Date of Assessment

2.Person providing responses: parents=1, other caregivers=2, Attendance=3

3.Difficulties describe by the family members:

Limb affected:

4.Right arm Involved =1 Not involved =2

5.Left arm Involved =1 Not involved =2

6.Right leg Involved =1 Not involved =2

7.Left leg Involved =1 Not involved =2

Gross motor problems: (GMF) simplified

8.• Sitting: without any support = 1/ with support = 2/ unable to do =3

9. • Toileting and Bathing: (without any support =1 / need help =2 / unable todo =3)

10. Any deformity/Contructure: Yes=1, No=2

20: GMFCS

1

2

3

4

5

11. Treatment needed :(Exercise =1/ Counseling =2/ Assistive device =3/Play=4, others=5Exercise =1

Counseling =2

Assistive

device =3

Play=4

others=5

12. Treatment provided :(Exercise =1/ Counseling =2/

Assistive device =3/Play=4/ others=5, referred to others =5

Exercise =1

Counselin

g =2

Assistive

device =3/

Play=4

others=5

referred to others =5

Assistive device: (Corner chair =1/ Wheel chair =2/ Special seating chair

=3/ Walking frame =4/ others =5)

13. Referral: To other hospital (for investigation/ other service)

=1, For education/special education =2. For vocational training=3

14. Comments of the therapist:

Appendix 12.6.3 Protocol for the social worker administering the insight strategies for visualsupport

1. LGA:

2. Name of child

3. Research No:

4. Phone number of Parents/Carers

5. How did you find the questions in the Insight history inventory?

A: 1. easy 2. Difficult

B: 1. Useful 2. Not useful

C: 1. Relevant 2. Irrelevant

8. Did you identify areas your child has problem with from the inventory?

Let us go through the areas together….(The social workers goes through questions thatreflect always or often from previous interview).

9. Which questions gave you the most concern for your child that you want treatment for?

10. Can we choose 8 of them to address?

Choose 8 questions that give concern and read out the strategies for all 8.

11. Can we choose the strategies you would like to start with:

Strategy 1:_____

Strategy 2:_____

Strategy 3:_____

Strategy 4:_____

Strategy 5:_____

Strategy 6:_____

Strategy 7:_____

Strategy 8:_____

13. Tell about the strategies being implemented at least three times a day

14. When would you like to start?_____

15. Do you think you will have problems administering the strategies? A.) Yes B.) No

16. What problems do you expect?.....
1…….

2……

3……

17. Suggest ways to help the patient and document your

recommendations (Page behind)Thank parent and let them know we

will call every two weeks to assess progress.

18. List the order in which you would like to implement the strategies.

Strategy 1:_____

Strategy 2:_____

Strategy 3:_____

Strategy 4:_____

Strategy 5:_____

Strategy 6:_____

Strategy 7:_____

Strategy 8:_____

19. How can we help you to implement these strategies??

20. Say good bye but remind on the importance of keeping the strategy document ready

21. Date of next follow up call……..

22. Time of phone call: 1. Afternoon 2. Morning 3. Evening

Explained in what language??.....

Appendix 12.7

Weekly follow up protocol for

social workers useFollow up 1

Phone call No……..

Greet and ask about the child or strategy doc:

1. Date of phone call:

2. Name of child

3. Research No:

4. Phone call answered: 1. Yes 2. No 3. Busy 4. Not going through

5. Phone call: Who are you speaking with?.....

6. Repeat call: 1. Yes 2. No……. Date:……..

Greet.

1. Were you able to implement the strategy A. Yes NO.

2. Were you able to do it 3 times daily A)

Yes

B) NOIf yes, skip to question 5

3. Why NO?

4.. If No? How many times were you able to do it?.....

Skip to question 6

5. If yes, How is the child coping with the first strategy?

6. Can we move to the next two strategies? A) Yes B) NO

7. If No, Why?

8. If yes, what strategies are next: 1. …….. and 2. ……..

We read and discuss the next two strategies as planned. Social worker asks if its understood.

9. If yes, proceed to fix another date for a phone call

Date of next phone call: Time:

10. If No the social worker repeats the strategy and explains it to the parent/carer.

Appendix 12.8: Demographic characteristics of children referred by

key informants anddiagnosis as non-cerebral palsy

Demographic characteristics of children referred by key informants and diagnosis as non-cerebral palsy			
		N	%
Total		* 383	11.7
Age	Mean (SD)	8.98	4.32
	Male	203	59.2
	Female	136	39.7
	Unknown	4	1.2
Infections			
	Post meningitis sequelae	6	1.7
	Polio	1	0.3

Neurologic/sensory problems			
	Intellectual difficulty	53	15.5
	Speech impairment	76	22.2
	Epilepsy	35	10.2
	Hearing impairment	42	12.2
	Behavioural disorder	7	2
	Squint	3	0.9
	visual impairment	2	0.6
	Down syndrome	102	29.7

Congenital deformities			
	Club foot	33	9.6
	Microcephaly	3	0.9
	Congenital multiple limb malfunction	3	0.9
	Congenital equino virus	2	0.6
	Congenital knee amputation	1	0.3
	Mesomelia	1	0.3
Heamoglobinopathies			
	Sickle cell anaemia	7	2
Nutrition			
	Rickets	3	0.9
Trauma			
	Spinal cord injury	1	0.3
	Injection palsy	1	0.3
	Fracture of the bone	1	0.3

- 343 children were examined, but some children had multiple associated comorbiditiesand more than one diagnosis (speech, hearing, intellectual difficulty and visual impairments) therefore the total number of diagnosis exceeds the number of children seen.

Appendix 12.9: Distribution of children that failed PVD tests

Number of PVD tests failed	Freq.	Percent	Cum.
0	86	22.16	22.16
1	21	5.41	27.58
2	24	6.19	33.76
3	20	5.15	38.92
4	14	3.61	42.53
5	23	5.93	48.45
6	23	5.93	54.38
7	15	3.87	58.25
8	23	5.93	64.18
9	14	3.61	67.78
10	25	6.44	74.23
11	40	10.31	84.54
12	60	15.46	100