Study Protocol: Longitudinal cohort study of oropharyngeal dysphagia: relationships to gross motor attainment, growth and nutritional status in preschool children with cerebral palsy

Journal:	*BMJ Open*
Manuscript ID:	bmjopen-2012-001460
Article Type:	Protocol
Date Submitted by the Author:	11-May-2012
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Primary Subject Heading:	Paediatrics
Secondary Subject Heading:	Nutrition and metabolism, Neurology
Keywords:	oropharyngeal dyphagia, deglutition disorders, cerebral palsy, gross motor function, Developmental neurology & neurodisability < PAEDIATRICS, NUTRITION & DIETETICS

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Short Title: Oropharyngeal dysphagia, gross motor, growth and nutrition in children with CP

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Key words: oropharyngeal dysphagia, deglutition disorders, cerebral palsy, gross motor function, growth, nutrition

Word Count:
Abstract: 302
Manuscript: 7005
References: 70
Tables/ figures: 3

ABSTRACT

Introduction

The prevalence of oropharyngeal dysphagia (OPD) in children with cerebral palsy (CP) is estimated to be between 19% and 99%. OPD can impact on children's growth, nutrition, and overall health. Despite the growing recognition of the extent and significance of health issues relating to OPD in children with CP, a lack of knowledge of its profile in this sub-population remains. This study aims to investigate the relationship between OPD, attainment of gross motor skills, growth and nutritional status in young children with CP at and between two crucial age points, 18-24 months and 36 months, corrected age.

Methods & analysis

This prospective longitudinal population-based study aims to recruit a total of 200 children with CP born in Queensland, Australia between 1st September 2006 and 31st December 2009 (60 per birth year). Outcomes include clinically assessed OPD (Schedule for Oral Motor Assessment, Dysphagia Disorders Survey, Pre-Speech Assessment Scale, signs suggestive of pharyngeal phase impairment, Thomas-Stonell and Greenberg Saliva Severity Scale), parent-reported OPD on a feeding questionnaire, gross motor skills (Gross Motor Function Measure, Gross Motor Function Classification System and motor type), growth and nutritional status (linear growth, body composition), and dietary intake (three day food record). The strength of relationship between outcome and exposure variables will be analysed using regression modelling with odds ratios and relative risk ratios.

Ethics and dissemination

This protocol describes a study that provides the first large population-based study of OPD in a representative sample of preschool children with CP, using direct clinical assessment. Ethics has been obtained through the University of Queensland Medical Research Ethics Committee, the Children's Health Services District Ethics Committee, and at other regional

and organisational ethics committees. Results are planned to be disseminated in six papers submitted to peer reviewed journals, and presentations at relevant international conferences.

ANZTR Trial Registration Number: ACTRN12611000616976

INTRODUCTION

Children with cerebral palsy (CP) may have poor feeding skills, influencing their growth, nutrition, and overall health[1, 2]. Cerebral palsy is the most common cause of physical disability in childhood, estimated at 2 per 1000 live born infants within Australia[3]. Cerebral palsy is an umbrella term which describes a group of disorders of movement and/ or posture and motor function, which is permanent but not unchanging and due to a non-progressive interference/ lesion in the developing brain[4]. Individuals with CP are an heterogeneous group, varying by severity and extent of motor involvement, type of movement patterns, aetiology, and related conditions[3].

The neurological lesion associated with CP may impact on the muscles of the jaw, cheeks, lips, tongue, palate and pharynx[5], which manifest functionally as difficulties with controlling saliva, eating, drinking, swallowing and speaking. Eating and drinking are complex sensorimotor activities, which can be described in four phases, including the oral-preparatory, oral (propulsive), pharyngeal and oesophageal phases of the swallow[6]. This study will focus on oropharyngeal dysphagia (OPD) in young children with CP, defined as impairment to any component of the oral and/ or pharyngeal phases associated with eating, drinking, or controlling saliva.

The oral-preparatory phase is initiated when food/fluid is taken into the mouth, and involves tasks necessary in bolus formation, including sucking, munching and chewing. Food and fluid are contained in the oral cavity surrounded by the upper dental arch and closure of the lips. Posterior leakage of the fluid bolus is prevented by contact between the soft palate and tongue, however this contact is not maintained during the processing of the solid food bolus. The oral (propulsive) phase involves the backward propulsion of the food bolus, by the tongue gradually expanding its contact with the hard palate posteriorly, to initiate the

pharyngeal swallow[6, 7]. The duration and movements necessary for the oral phases differ depending on the child's age and the utensils used to ingest food/ fluid[7]. The oral-preparatory phase of the swallow also differs when ingesting food compared to fluid boluses. When defining the swallow stages for solid foods, Matsuo and Palmer advocate the use of the Process Model of Feeding, because of the overlap between the phases described in the Four Stage/ Phase Model for fluids[6]. The Process Model divides the oral-preparatory phase into Stage I Transport and Food Processing, in which the food is first ingested and moved onto the lateral occlusal surfaces of the teeth before being masticated to an optimal consistency for swallowing.

The pharyngeal phase is used to describe the passage of both food and fluid boluses through the pharynx, although when ingesting fluids it normally overlaps with the oral propulsive phase[6]. On initiation of the pharyngeal phase, the soft palate elevates to seal the nasopharynx to prevent nasal regurgitation. The tongue base retracts, propelling the bolus posteriorly against the pharyngeal walls followed by the pharyngeal constrictor muscles contracting to squeeze the bolus downward. To ensure airway safety during bolus passage, respiration ceases momentarily, the vocal folds close, the arytenoids tilt forward to contact the base of the epiglottis, the larynx elevates under the base of the tongue and the epiglottis tilts backward to seal the laryngeal vestibule. The opening of the upper oesophageal sphincter (UOS) is facilitated through the relaxation of the cricopharyngeous muscle, contraction of the suprahyoid and thyrohyoid muscles, and the pressure of the descending bolus[6]. The oesophageal phase is the final phase of the swallow, which begins as the bolus moves through the UOS, to be transported via automatic peristaltic waves to the stomach[7].

Specific patterns of oral, pharyngeal, and oesophageal impairments in feeding have been documented in children with CP. They may have difficulty in the oral phase of the swallow

due to inadequate function of the oral muscles, exaggerated oral reflexes and altered oral sensitivity[8]. This may include limitations to tongue lateralisation necessary for chewing solids, excessive tongue thrusting, impaired bolus transit, increased oral transit time (greater than three seconds), and reduced ability to clear food residue in the mouth. Poor control of the lips may result in difficulty receiving the bolus (eg sipping from a cup or clearing a spoon), difficulty sucking from a bottle or straw, anterior loss of food due to poor lip seal, and excessive saliva loss[8]. Children may also have pharyngeal phase impairments, including delayed or incomplete closure of the airway during the swallow, oropharyngeal aspiration of food or fluid, and food residue in the pharynx[9]. Aspiration is defined as passage of material below the vocal folds[6]. This can be oropharyngeal aspiration (primary) of orally ingested material, saliva or mucous secretions; or reflux aspiration (secondary) of gastro-oesophageal refluxate. Aspiration can occur before the swallow (due to lingual disco-ordination allowing the bolus to prematurely spill over the base of the tongue, or a delayed swallow trigger); during the swallow (associated with ineffective laryngeal closure or disco-ordination); or after the swallow (related to laryngeal/ pharyngeal residue falling into the reopened airway)[6]. Usually food entering the laryngeal vestibule and subglottic space triggers a cough, which is a major protective mechanism of the airway[6]. Silent aspiration occurs when food or fluid enters below the true vocal folds with the absence of clinical signs or symptoms, which is commonly reported in children with CP[9, 10]. Gastro-intestinal impairments (including reduced motility and reflux) occur frequently in individuals with feeding problems and CP, both secondary to and contributing towards the difficulty[11].

It is believed OPD is highly prevalent in individuals with CP, however there is a lack of comprehensive population-based data[5, 12-26]. Estimates of prevalence vary significantly, from 19% in a large register sample[24], to 99% in a sample of children with moderate-severe gross motor impairment[14]. Much of the literature exploring OPD in feeding has

been limited by study methodology and case-definition of OPD. Many studies have based the prevalence of OPD on parent report or non-validated methods, and samples have generally been limited to individuals with more severe gross motor impairments[12, 14, 15, 17] and across a broad age range[5, 12, 14-19, 21, 22, 26]. The findings from key studies have been summarised in Table 1.

Indirect or inconsistent means of OPD case identification have regularly been utilised in studies, with OPD identified through parent report[12, 13, 15-17], chart reviews[5, 17], and non-standardised assessments[21-23, 25]. The variability in the method of case identification limits comparisons between these studies, and makes it difficult to estimate the true prevalence of OPD in the paediatric CP population. Parents have been shown to underestimate the presence of impaired feeding skills compared to formal clinical evaluation[14], so prevalence data using these methods may represent an underestimate of the true population prevalence of OPD. Most parent questionnaires in the reported studies lacked adequate validity and reliability data, reducing confidence in these results[12, 13, 15-17].

The generalisability of prevalence estimates of OPD to the general population of children with CP has been limited in most studies due to a focus on feeding skills in children with moderate-severe gross motor impairment[12, 14, 15, 17, 25]. Many of the studies which sampled across the range of gross motor severity have still had a disproportionate number of

Table 1 Prevalence of Oropharyngeal Dysphagia in Children with Cerebral Palsy and its Relationship to Gross Motor Function

Author & Year	Participants	OPD Measure	Gross Motor Measure	Major Findings
Santoro, et al. (2011)	n=40 children with CP and feeding problems aged 4mths-11 years, GMFCS III-V	Parent questionnaire and mealtime observation by SP	GMFCS CP motor type	Children from GMFCS III showed best feeding performance (hemi/ diplegic CP)
Erkin, et al. (2010)	n=120 children with CP, 2-18 years	Informal observations of feeding behaviours	GMFCS (collapsed to 2 grps) CP motor type	22% feeding dysfunction (12% mild, 8% moderate, 2% severe). Feeding dysfunction in 4% of GMFCS I-III, and 22% of GMFCS IV-V ($p<0.001$)
Parkes, et al. (2010)	n=1357 children with CP, median 5;11 yrs, GMFCS I-V	Question on standardised assessment for register ('absent' or 'present')	GMFCS CP motor type (Surveillance of CP in Europe Project)	19% chewing and swallowing problems GMFCS significantly related to swallowing/ chewing difficulties and excessive drooling: GMFCS IV – OR 4.8 GMFCS V – OR 15.7
Wilson & Hustad (2009)	n=37 children with CP, 11-58 mths (mean 41 mths)	Parent report on feeding and swallowing Questionnaire Clinical evaluation of OPD (no formal tools)	No analysis of motor severity	56% had difficulty feeding from a bottle 78% had oral motor involvement (including motor speech) No analysis with gross motor
Ortega, et al. (2009)	n=53 children with CP, 3-13 yrs, GMFCS I-V (with 75% of sample from IV-V)	Oral Motor Assessment Scale	GMFCS	83% did not have functional feeding skills No analysis with gross motor
Calis, et al. (2008)	n=166 children with severe CP and ID, 2-19 yrs (mean 9;4 yrs). GMFCS IV-V, IQ<55	DDS and DSS Parent report	GMFCS	99% clinically apparent dysphagia Oral motor severity positively associated with motor functional severity ($p<0.001$) Postural stability positive association to DDS score, but not postural alignment for eating.
Yilmaz, et al. (2004)	n=23 children with spastic CP, 4-25 years GMFCS I-V	FFAm	Ambulatory status	50-74% normal-mild feeding difficulties; 30-51% moderate-severe feeding difficulties
Field, et al. (2003)	n=44 children with CP, 1 mth -12 yrs (median age range 13-36 mths)	Record review	No analysis of motor severity	68% oral motor delay 32% dysphagia
Fung, et al. (2002)	n=230 children with CP, 2-18 yrs (mean 9.7yrs), GMFCS III-V	Parent reported on feeding questionnaire – rated as none, mild, mod,	GMFCS	48% feeding problems. GMFCS level was highly associated with the degree of feeding dysfunction ($p<0.001$).

		severe		
Sullivan, et al. (2000)	n=271 parents of children with childhood impairments (96% CP), 4-13 yrs, mild-severe gross motor	Register question to determine 'articulation/ swallowing problems'. Parent questionnaire to investigate specific feeding problems.	Parent rated severity of motor function, relating to aids needed (mild, mod, severe)	79% articulation or swallowing problems. Significant correlation between severity of gross motor impairment and range of specific feeding problems (eg choking with food $p<0.001$; prolonged mealtime $p<0.001$).
Reilly, et al. (1996)	n=49 children with CP, 12-72mths, mild-profound (70% with severe-profound imp)	SOMA Early feeding histories	Standard Recording of Central Motor Deficit – classified as no disorder/ mild; severe/ profound.	Positive relationship between OPD severity and gross motor severity ($p=0.000$) Mod and severe OPD more common in tetraplegia, whereas diplegia was associated with mild OPD ($p=0.001$).
Dahl, et al. (1996)	n=35 children with CP, 2.4-15.2yrs (mean 7.7yrs), profound motor handicaps (moderate and severe CP)	Parent interview (retrospective data of 4 weeks) triangulated with medical file review	Motor severity differentiated by level of dependence	60% reported as having daily feeding problems No analysis of gross motor
Stallings, et al. (1993)	n=142 children with quadriplegic CP, 2-18 yrs.	Parent interview (0-5; 0=no problems, 5=all (5) oral motor problems)	Diagnostic criteria (for quadriplegic CP) not defined in paper	86% impaired oral motor ability No analysis of gross motor
Waterman, et al. (1992)	n=56 children with CP, 5-21yrs (median 14 yrs), mild-severe	Chart review (clinical or radiographical dysphagia) Interviews with SP	Severity defined based on ambulatory status from chart review	27% had evidence of swallowing disorders More severe CP in dysphagic group ('consistent but non-significant trend'– no statistics reported)
Thommessen, et al. (1991)	n=42 children with CP, 1-16yrs	OPD evaluated by 3 OTs/PTs (based on child's age)	No analysis of motor severity	33% had OPD No analysis
Love, et al. (1980)	n=60 children with CP, 3-23yrs (mean 12.5yrs), spastic, athetoid and mixed; mild – non-ambulatory	Non-standardised oral-motor tasks (biting, sucking, swallowing, chewing soft and firm food)	No analysis of motor severity	40% with inadequate feeding

Key: CP Cerebral Palsy; DDS Dysphagia Disorders Survey; DSS Dysphagia Severity Scale; FFAm Functional Feeding Assessment modified; GMFCS Gross Motor Function Classification System; ID Intellectual Disability; imp impairment; IQ Intelligence Quotient; mod moderate; mths months; OPD Oropharyngeal dysphagia; OR Odds Ratio; OT Occupational Therapist; PT Physiotherapist; SOMA Schedule for Oral Motor Assessment; SP Speech Pathologist; yrs years

individuals from the more severe classifications[5, 16, 18, 20, 21]. This is largely due to sampling bias, with most studies recruiting from special schools or clinic databases, and thus limiting the sample representativeness. In addition, a range of measures have been used to determine gross motor severity, including formal classification systems such as the Gross Motor Function Classification System (GMFCS), and criteria developed for the individual study. This limits our ability to accurately quantify the prevalence of OPD across the full range of gross motor severity, from mild to severe, and may provide an overestimate of the prevalence in the general population of children with CP if rates are extrapolated based on the moderate-severe sample.

Feeding skills develop rapidly in the early years as children transition through a range of food and fluid textures, related to their developing anatomy, neurology and physiology[27]. Rapid development of sensorimotor integration of swallowing and respiration, upper limb skills, posture and psychosocial maturation occur during the first three years[7]. By 18 months children are typically sitting independently, with fully co-ordinated swallow and respiration, and taking a full range of textures[7]. The development of chewing skills continues into childhood, with the adult co-ordination of lateral and vertical jaw movements emerging between three and six years[28]. Most prevalence studies of OPD in children with CP have been designed to examine oral sensorimotor skills in samples with a broad age range from early childhood (four months to four years) through to adolescence or early adulthood (11-25 years)[5, 12, 14-19, 21, 26]. The mean age for many of these studies was nine years. Only two studies limited their sample to preschool years, with participants ranging in age from 12-72 months[20] and 11-52 months[13]. Few children from the toddler or preschool age range have been sampled in previous studies, so a gap in knowledge remains. It is important to begin to delineate OPD in this critical age range to facilitate early identification and

intervention, and to explore the progression of early feeding skills and their changing relationships with other associated factors (e.g., growth, nutrition and respiratory health).

It is well accepted clinically that there is an interaction between an individual's oral sensorimotor skills in feeding and their gross motor skills. An individual's feeding posture can impact on their swallow by promoting poor alignment or reducing the stability for controlled oral movements, as well as the influence of the neurological lesion on all motor skills[15, 29]. Poor head position has been related to compromised airway protection by opening the airway, and influencing the flow rate of foods/ fluids swallowed[30]. The precise relationship between body position and swallow-breath coordination continues to be explored[31]. This relationship between OPD and gross motor skills is supported in the literature, with the prevalence and severity of OPD reported to be positively correlated with the extent of motor involvement[5, 14-16, 20, 24]. However, these findings lack weight due to few studies using direct objective measures of oral sensorimotor skills[5, 14-16, 23, 24], a lack of validated measures of gross motor skills[5, 16], or sampling only children with moderate-severe gross motor impairment[14, 15, 25].

The Oxford Feeding Study of 271 children with OPD, found those with more extensive motor involvement, i.e. quadriplegia and dyskinesia, were most likely to have difficulties with swallowing and articulation, based on parent report[16]. Those unable to walk or who required an aid and helper to walk were more likely to have problems eating and swallowing lumpy food, to need food mashed or liquidised, and were also more likely to be fed via a tube. In a large register-based study (n=1357), the odds of having swallowing/ chewing difficulties and excessive drooling increased significantly as GMFCS level increased[24], however this study only used a single standardised question to determine presence of feeding

difficulty. Using validated assessments (Schedule for Oral Motor Assessment and Standard Recording of Central Motor Deficit categories) the presence of gross motor impairment was significantly associated with the presence of oral motor dysfunction in a cross-sectional community-based sample of 49 preschool children with CP[20]. While strengthened by using validated measures for both oral motor and gross motor skills, the sample was small, and only used binary outcomes (presence/ absence of dysfunction). The relationship between OPD and gross motor skill attainment will be strengthened by exploring this association across a number of gross motor severity levels using the GMFCS.

The feeding impairments resulting from OPD may impact negatively on many dimensions of an individual's health, including the child's development, growth and nutrition, chest status and respiratory health, gastro-intestinal functioning, and parent-child interactions[32]. Both OPD and tube feeding are demonstrated risk factors for increased premature mortality in individuals with CP[33-35]. Optimal nutrition in the early years forms a critical foundation for improved health across the lifespan. Compromised nutritional status influences children's mood and irritability, muscle spasticity, healing, peripheral circulation and general well-being [36]. In addition, OPD can result in acute and/ or chronic oropharyngeal aspiration which is significantly associated with compromised respiratory status, including recurrent lower respiratory tract infections and chronic lung disease[9, 19]. Understanding the nature and severity of OPD in young children with CP and its relationship to gross motor attainment, growth and nutrition, will inform health interventions, benefiting children with CP and their families, and potentially lowering costs of health care[37].

Aims and hypotheses

This study will investigate the relationship between OPD, gross motor skills, growth and nutritional status in young children with CP across two critical age points, 18-24 months and 36 months, corrected age. Specifically, this study aims to:

1 i) Systematically review the literature determining the clinimetrics of measures of OPD in preschool children with CP.

ii) Test the psychometric properties of the Schedule for Oral Motor Assessment (SOMA), Dysphagia Disorders Schedule (DDS) and Pre-Speech Assessment Scale (PSAS) in young children with CP.

2 i) Determine the prevalence of OPD and its subtypes (impaired saliva control, oral phase impairment, and pharyngeal phase impairment) in a population of children with CP at 18-36 months.

ii) Explore the nature of the relationship between OPD and gross motor functional severity (according to GMFCS levels); and growth and nutritional status.

3 Longitudinally examine the potential risk factors for OPD (including gross motor attainment, anthropometric measures, dietary intake, ingestion functions, food and fluid textures, gender, age, socio-economic factors) in children aged 18-24 and 36 months with CP.

These aims will be explored through the following three hypotheses:

H_1 The SOMA and DDS will be the most valid and reliable measures of OPD in young children with CP. The PSAS will have the best clinical utility.

H_2 (i) There will be a negative relationship between OPD prevalence and gross motor function in children with CP aged 18-36 months.

(ii) There will be a positive relationship between OPD, poor growth and nutritional status in children with CP aged 18-36 months.

H3 Gross motor function, poor growth and nutritional status will have a greater association with OPD in children with CP than demographic risk factors.

Study significance

The results of this study will:

- Determine the accuracy of the SOMA, DDS, PSAS and signs suggestive of pharyngeal phase impairment, in detecting and evaluating OPD in preschool aged children with CP.
- Contribute population-based data on the prevalence of OPD and subtypes, in children with CP using standardised measures. To date there is limited comprehensive population data across all gross motor severity levels. This data is essential before intervention trials can be conducted.
- Delineate the relationship between OPD and gross motor skill attainment in children with CP. Greater understanding of this relationship will assist in proactive screening in early intervention services, including early detection of children at risk of aspiration and compromised chest status, and prevention of negative health effects.
- Further explore potential associations between OPD and nutritional status and growth in children with CP. This will allow for greater access to preventative nutritional treatments and the development of more targeted interventions, thus promoting growth and overall health outcomes in young children with CP.

METHODS AND ANALYSES

This prospective longitudinal cohort study aims to recruit 200 children with CP born in Queensland, Australia, between 1st September 2006 and 31st December 2009. The OPD study is part of a larger longitudinal population-based study, Queensland CP Child: Growth, Nutrition and Physical Activity, which is exploring growth, nutrition and physical activity in

children with CP (National Health and Medical Research Council (NHMRC) Australia, 569605). This study is being conducted in conjunction with another study, Queensland CP Child: Motor Function and Brain Development Study (NHMRC 465128). Figure 1 visually represents the relationship between these studies and the OPD sub-studies, which include:

1. Validity and reproducibility studies
    a. Discriminative validity with typically developing reference sample
    b. Convergent validity with an additional OPD measure
    c. Reproducibility (test-retest, intra-rater, inter-rater)
2. Cross-sectional study of children aged 18-36 months
    a. Overall prevalence of OPD, subtypes and association with gross motor
    b. Oral phase impairment
    c. Pharyngeal phase impairment
    d. Functional feeding skills on food and fluid textures
3. Longitudinal study of children between 18-24 months and 36 months

Recruitment

State-wide subject recruitment commenced in July 2007 in collaboration with the Queensland Cerebral Palsy Register, the Queensland Cerebral Palsy League, the Royal Children's Hospital (RCH) Brisbane, the Queensland Cerebral Palsy Health Service, the Royal Women's Hospital Brisbane, and the Mater Children's Hospital. Paediatricians, general practitioners, allied health professionals, child health nurses, and neonatal follow-up clinics are encouraged to refer children with motor delay (not sitting at 10 months, not standing at 12 months or walking at 24 months) for confirmation of a diagnosis of CP at the RCH/Mater Mothers' Hospital Specialist clinics. High ascertainment is expected for children across all levels of motor severity (GMFCS I to V) particularly as many of these children access

services through the Queensland Cerebral Palsy Health Service, one of the key referral sources. Children who are detected after 18 months of age will be entered into the study later, at the time of diagnosis. Children can enter the study at 18, 24, 30 or 36 month age points. Those entering at 18 or 24 months will have their second assessment point collected at 36 months, and will be included as part of the longitudinal study. Children entering at 30 or 36 months will have their second assessment at 48 months, therefore will not be included in the longitudinal study detailed in this study protocol. Further details of study entry and feasibility can be found in the larger study's protocol[38].

Forty children with typical development aged 18-36 months (stratified for age) will be recruited to participate as a reference sample for the study. Siblings of children participating in the overall study will be invited to participate, as well as recruitment through staff newsletters, a hospital childcare centre, and participants from other studies within the centre.

Selection Criteria

Inclusion Criteria

Children aged 18-36 months corrected age at the time of evaluation (birth years 2006-2009), born in Queensland, with a confirmed diagnosis of cerebral palsy are invited to participate in the present study. For the present study, CP is defined as a disorder of movement and/ or posture and motor function, which must be permanent but not unchanging, and due to a non-progressive interference/ lesion in the developing brain (congenital lesions only)[4]. The characteristic motor types are spasticity and dyskinesias (ataxia, rigidity and dystonia), and clinical features may also include negative signs of the motor neurone syndrome (muscle weakness and poor selective motor control)[39].

Exclusion Criteria

Children diagnosed with a progressive or neurodegenerative lesion and children born outside Queensland are excluded from the study.

Typically Developing Reference Sample:

Children are eligible to participate in the reference sample if they are aged 18-36 months; born full term (<37 weeks); with no admissions to neonatal care, no diagnosis receiving medical or allied health care; and not on regular medications.

Measurements and Procedures

Following confirmation of a diagnosis of CP, children attend the RCH for an assessment session with their family. During this visit, children are assessed using the Gross Motor Function Measure (GMFM)[40], Manual Ability Classification System (MACs) [41], anthropometric measurements taken, questionnaires administered to the parent/ caregiver verbally (including the Pediatric Evaluation of Disability Inventory[42], and Queensland CP Child: Growth, Nutrition and Physical Activity: Feeding Questionnaire) and the child's mealtime is videotaped.

Children participating in the reproducibility study will be invited to return to the hospital within a month to have a repeat mealtime video. If this is not possible, a home visit will be conducted. Children participating in the typically developing reference sample will be assessed at the hospital or at home, for a single mealtime video.

Feeding Evaluation

During the feeding assessment, the child is well positioned in their typical mealtime seating (ie chair, stroller, carer's arms). The video camera is set up to include a view of the child's face and neck, angled to the side of the feeder's shoulder of the hand that is not feeding the child, as per the study snack protocol. Prior to and following the mealtime, the researcher videoing the session records observations regarding clinical swallow signs (wet/ gurgly voice, wet/ gurgly breathing, rattly chest, or presence of cough) and severity of drooling. These ratings are confirmed by the speech pathologist when rating the videos. During the video session, the child is given three standardised presentations of each of four textures (puree, lumpy, chewable and fluid) by their primary carer, as outlined in the Schedule for Oral Motor Assessment (SOMA) administration manual[43]. Purees include foods such as yoghurt, mousse or pureed fruit. Lumpy foods could include semi-solid (e.g., baked beans, roughly mashed vegetables) or solid foods (e.g., fruit salad) from a spoon. For the purpose of this assessment, chewable foods are items that are finger fed, usually requiring biting, including biscuits or whole fruit. Following these standard presentations, the child is allowed to complete the snack eating independently or assisted by their primary carer.

Primary Measures

A major limitation in studies of OPD is the lack of widely accepted, validated and reliable measures[15]. The aim of the present study is to gather information regarding OPD which reflects children's performance in naturalistic environments (eg. home and childcare centres). For this reason, non-invasive observational methods were selected as part of the standard protocol for all children. The Schedule for Oral Motor Assessment (SOMA)[50], Dysphagia Disorders Survey: Paediatric (DDS)[51], and Pre-Speech Assessment Scale (PSAS)[44] were selected through systematic review as the most appropriate non-invasive objective clinical measures for the detection of dysphagia for this study[45]. The video tapes of children's

mealtimes are formally rated by an independent speech pathologist, and data recorded using the standard assessment forms. Sixteen clinical pharyngeal signs suggestive of aspiration are also rated for each food/ fluid texture, in conjunction with the rating completed in the session. The use of videos in mealtime observations is recommended in the SOMA administration manual to allow repeated viewing for more accurate description of motor tasks. The speech pathologist is certified in the use of the DDS to meet the validation standards[46]. The allocation of GMFCS level is masked to the speech pathologist when rating mealtime videos. If clinically indicated, some children have further evaluation of their OPD using instrumental assessments, such as Video Fluoroscopic Swallow Study (VFSS). This information is collected when available but is not part of the standard protocol for all children.

Schedule for Oral Motor Assessment (SOMA)

The SOMA is a standardised discriminative assessment which quantifies OPD in children aged between 8-24 months[43]. It was originally designed to evaluate children with no/ mild neurological dysfunction, but subsequently was used to evaluate oral motor dysfunction (OMD) associated with a number of causes including neurological impairments[43]. The tool categorises children as OMD or normal oral motor function based on specified thresholds for each of seven oral motor challenge categories (OMCC) (puree, semi-solid, solid, cracker, bottle, trainer cup, cup)[43]. The tool is predominantly a test of oral phase dysfunction, however some items pertain to swallowing and the pharyngeal phase. Children are only scored on food/ fluid textures they accept during the assessment. The standardised administration of textures outlined in the administration manual is maintained in this study as much as possible, while allowing some flexibility for individual child and family factors to optimise the naturalistic context of the assessment.

The SOMA has been validated on 127 young infants; 58 comparison children with typical oral skills, 56 with non-organic failure to thrive (aged 8-24 months), and 13 children with CP and overt feeding difficulties (aged up to 42 months)[47]. The abnormality score (total number of OMCCs with OMD) for children with CP was significantly different from the comparison group ($p<0.0001$). Individual OMCCs do not have adequate discriminative validity reported to be analysed as individual subtests, with 8-77% false negatives in the CP group[47]. The reliability of the measure was established by two independent speech pathologists rating three trials of ten randomly selected videos from the sample. It has strong inter-rater reliability ($\kappa=1.0$ in 68% of fluid category items and 58% of food category items) and test-retest reliability between boluses ($\kappa=1.0$ in 84% of items)[48].

Dysphagia Disorders Survey (DDS)

The DDS was developed as an evaluative screening tool to assess feeding and swallowing function in children and adults with a developmental disability[46]. Through observation of a typical mealtime, it identifies those with signs of oral preparation, oral initiation, pharyngeal and oesophageal phase dysphagia[46]. The measure is divided into two distinct parts: Part 1 scores dysphagia related consequences (such as low weight, adaptive utensils and position); Part 2 rates the specific oral functions observed across three textures (non-chewable food, chewable food and fluid). The raw score from Part 1, and percentiles which are derived from both Part 1 and 2, are not used in this study as they assess consequences of mealtime difficulty rather than specifically OPD. Part 2 provides a raw score that indicates an individual's functional eating competency (with a maximum impairment raw score of 22) and has been used previously as a measure of OPD[14]. The standardised scoring of the DDS has been modified for this study to rate any atypical tasks (whether functional or not) as impaired (OPD).

The DDS underwent final standardisation on 427 individuals with mean age 33 years[14]. The paediatric measure was developed in a group of 166 children (range 2 years 1 month-19 years 1 month; mean 9 years 4 months), with moderate-severe CP (GMFCS III-V) and intellectual disability[14]. Test validity and inter-item reliability were derived from an initial sample of 626 people with developmental disability[49]. Convergent validity was demonstrated in two studies comparing DDS scores to blinded speech pathologist diagnosis[14, 46]. Inter-rater reliability of 97% agreement was calculated from a sample of 21 participants rated by six speech pathologists (each pair of speech pathologists rated seven participants)[46].

Dysphagia Severity Scale (DSS)

The DSS was developed by Calis et al. (2008) to provide a severity rating from the DDS Part 2 raw scores[14]. Individuals are classified as one of four severity levels, with level one being no disorder, and level four a profound disorder. The mild classification and moderate-severe classification are differentiated by the presence of pharyngeal phase impairments (items 13-14 on the DDS), in addition to a score of one or more on the DDS Part 2. A profound disorder is reflected by non-oral status of individuals due to the severity of their OPD.

Pre-Speech Assessment Scale

The PSAS is an evaluative measure that examines 27 pre-speech feeding behaviour performance areas related to sucking, swallowing, biting, chewing, respiration-phonation and sound play[44]. It is appropriate for use with children with a neurological impairment, as well as those with typical development. Each subtest is scored on an ordinal abnormality scale (1-9) and a developmental scale (with age norms to 24+ months), to provide a double score

overall. This provides comprehensive information on both dysfunctional and delayed feeding behavior expected up to 24+ months.

The PSAS was developed through a three year longitudinal study of six children, and field testing of the measure for eight years by 215 trained clinicians who provided annual feedback on its clinical use[44]. Other aspects of the measure's validity have not been tested. Reliability has been shown to be strong, although only in two studies with limited methodology[44, 50]. Intra-rater reliability was 96% for 25 feeding behaviours which were scored in the six typically developing children[50]. Inter-rater reliability for this same sample was similarly excellent between two raters (92%)[50]. Inter-rater reliability was fair to good when rated from video footage, with 65-87% agreement when 75 clinicians' ratings were compared to a pre-determined standard of correctness for 78 children[44].

Signs suggestive of pharyngeal phase impairment

Pre- and post-mealtime observations of the presence or absence of (i) wet/ gurgly voice (ii) wet/ gurgly breathing, (iii) rattly chest and (iv) cough are rated face-to-face in the mealtime session by a trained research assistant, to assess clinical signs of pharyngeal phase difficulty. A determination of pharyngeal phase impairment is noted if a child demonstrates any one of these signs, or one of 16 signs rated from video by the speech pathologist. These behaviours include gagging, coughing, choking, vomiting, throat clearing, multiple swallows, wheezing, stridor, rapid or laboured breathing, gurgly voice, rattly chest, snuffly nose, eye tearing, circumoral cyanosis/ duskiness, and food refusal, and are noted for each food and fluid texture. These signs were selected from the literature[10, 51] and research conducted by one of the investigators (KW)[52].

A cross-sectional study of 150 children with dysphagia (mean age 16 months) compared retrospective data of pharyngeal phase impairments identified by VFSS to 11 commonly reported clinical signs and symptoms to determine their sensitivity and specificity[52]. Wet voice (sensitivity 0.67, specificity 0.92), wet breathing (sensitivity 0.33, specificity 0.83) and cough (sensitivity 0.67, specificity 0.53) were considered good clinical markers of oropharyngeal aspiration on thin fluids, but not for puree textures.

Thomas-Stonell & Greenburg Scale – Saliva Control

The Thomas-Stonell & Greenberg (1988) scale is a semi-quantitative assessment of drooling severity (one to five point scale of no drooling to profuse drooling) and frequency (one to four point scale of no drooling to constant drooling)[53]. A pre- and post-mealtime severity rating is recorded by trained researchers within the mealtime assessment and confirmed by the speech pathologist from video. In addition, a severity and frequency rating by the parents is collected based on observations during the previous week, and information reporting on the representativeness of this rating.

In a case-control study of 14 children with saliva loss and spastic CP aged 7-18 years (mean 11;7 years), drooling frequency and severity were reported by parents on the Thomas-Stonell & Greenberg Scale[54]. A Drooling Quotient, derived from parent scores, was compared to a more objective measure of weighing saliva loss on bibs and shown to be positively correlated (Spearman's rho=0.604 $p<0.05$)[54].

Gross Motor Function Classification System (GMFCS)

The Gross Motor Function Classification System (GMFCS) is a five level classification system of children's functional gross motor severity. It is based on self-initiated movements,

anti-gravity postures and motor skills expected in a typical five year old[55]. Children who are independently ambulant are classified as GMFCS I or II, those requiring an assistive mobility device to walk classified as GMFCS III and those in wheeled mobility as GMFCS IV and V. Two physiotherapists, trained in the use of the GMFCS, independently observe and classify children in one of five functional categories[55].

The GMFCS has internationally established validity, reliability and stability for the classification and prediction of motor function of children with CP aged 2-12 years[55-57]. It has a high inter-rater reliability (generalisability=0.93)[56]. Classification of gross motor abilities change with age, therefore separate descriptions are used for different age bands. In the current study, the <2 years and 2-4 year descriptions are used. Lower inter-rater reliability is documented for the <2 years age band (κ=0.55), as younger children's gross motor abilities are more variable, and less developmental information is available on which to base the classification [58]. Test retest reliability from <2-12 years appeared to be acceptable (generalisability coefficient=0.68). The GMFCS has been correlated with a number of motor scales, as well as CP distribution and type of motor impairment[59].

Anthropometry

Height or length (depending on children's ability to stand) is measured to the last completed millimetre by a portable stadiometer/ length board (Shorr Productions, Maryland USA). Where a direct measure of height or length is not possible, height is estimated using published equations from knee length or upper arm length[60] measured with an anthropometer (Holtain Ltd, UK). Weight (measured to the nearest 100 grams using chair scales (Seca, Germany)) and skin-fold thickness (tricep and subscapular skinfolds, measured in millimetres with Harpenden callipers (Holtain ltd UK)) measures are taken and Body Mass

Index (BMI) calculated (as weight/ height $(m)^2$) to assess children's nutritional status. Skin-fold measurements and BMI will be converted to z scores for analysis[61]. All measures are conducted by trained investigators. Full details of anthropometric procedures are detailed in the larger study protocol paper[38].

Dietary Intake

A three day weighed food record is used to measure children's typical dietary intake[62]. Parents are instructed on the standard protocol to ensure accuracy and consistency in completing the food record. Food records will be analysed for the percentage of children's diet made up of food and fluid textures. Food records are also analysed using the Foodworks™ dietary analysis software program (Xyris Software (Australia) Pty Ltd) to give information regarding energy, carbohydrate, fat and protein intake.

Secondary Measures

Queensland Cerebral Palsy Child: Growth, Nutrition and Physical Activity: Feeding Questionnaire (Qld CP Child Feeding Questionnaire)

The Qld CP Child Feeding Questionnaire gathers parent report on their child's oral sensorimotor and mealtime function. Parent report will be used to triangulate findings from clinical assessment to gain a more comprehensive picture of the child's skills across settings and time. It includes:

- Severity and frequency of saliva loss using the Thomas-Stonell and Greenberg Scale (above).
- The impact of saliva on four domains, including the impact on child and family measured using a ten point visual analogue scale.

- Types of food and fluid included in the child's diet: inclusion of textures rated for four fluid levels (thin, mildly thick, moderately thick and extremely thick) and five food textures (puree, thick puree, lumpy mashed food, chewable solids and tough chewable foods). Fluid terms align with the Australian Standardised Labels and Definitions[63].
- Presence of eating or drinking problems: rated on four point scale from no feeding problems to severe difficulties. Severity is also rated for eating and drinking on a ten point visual analogue scale.
- Mealtime behaviours and signs suggestive of pharyngeal phase impairment or aspiration, are documented by parents against the same 16 signs and symptoms suggestive of pharyngeal phase impairment as is noted in clinical observation. Presence or absence of specific signs and symptoms were noted on each texture (thin fluid, thick fluid, puree, lumpy, finger foods).

Gross Motor Function

Gross Motor Function is evaluated at each assessment using the Gross Motor Function Measure (GMFM-66 and GMFM-88)[40]. The GMFM is an evaluative tool that covers five gross motor domains, including lying & rolling sitting; crawling & kneeling; standing; walking, running & jumping. The GMFM-66 is a subset of items from the GMFM-88, developed through Rasch analysis, and shown to be valid and reliable in children with CP[64]. The GMFM-66 will be used to provide an overall measure of gross motor function, and the GMFM-88 domain scores to explore specific motor skills. Scores are expressed as a percentage of the maximum score, which are skills expected of a typically developing child at five years [65]. The GMFM is not valid for comparisons of children across different age ranges, therefore all analyses using GMFM scores are completed 18-24 month and 30-36

month age brackets. Gross motor assessment is completed by two experienced paediatric physiotherapists who have criterion rating with the study developers (RB).

Motor Type and Distribution

The type of CP (spastic, dykinetic, hypotonic) and motor distribution (hemiplegia, diplegia, quadriplegia) is classified according to the Surveillance of CP in Europe[66]. This is assessed by two independent physiotherapists at each assessment.

Manual Ability Classification System (MACs)

Children's manual ability is classified during performance in everyday activities according to the MACs. The MACs classifies children on a five level scale based on how they use their hands when performing activities such as eating, dressing, playing and drawing[41]. This classification was developed for children aged 4-18 years, but has been shown to have good reliability for use in children as young as two years[67]. Children are rated by two independent physiotherapists.

Sample size calculations

Queensland Cerebral Palsy Child: Growth, Nutrition and Physical Activity

Based on a reported incidence of CP of 2/1000 live births within Australia, there is an estimated 100 new cases of CP in Queensland each year[3]. For sample size calculations, a population prevalence estimate of 90% was taken from the study by Reilly and colleagues[20]. In order to estimate the true prevalence of OPD in the population of children with CP with 95% confidence, a minimum sample of 35 participants were needed to provide sufficient precision within ±10% of the true value.

Due to the limited data reported in the literature of prevalence based on direct clinical evaluation in the mild gross motor level, children in GMFCS I were hypothesised to have normal feeding skills. Nearly all children in GMFCS V have been reported to have OPD[14]. With an expected 40 participants per GMFCS level (total n=200), this study will be able to detect a significant difference between groups (80% power, alpha=0.05) if the true proportion of OPD in the population differs by >25% between groups.

Validity and reproducibility studies

Oropharyngeal dysphagia reproducibility study

With an expected agreement of greater than 90%, a sample of 20 children with CP per age band (a total of 40 children across 18-36 months age range and gross motor severity levels) will be able to give sufficient statistical power, with 95% confidence.

Oropharyngeal dysphagia discriminative validity study

In order to estimate the true mean score of typically developing children aged 18-24 months and 30-36 months on the SOMA and DDS with 95% confidence (and precision of 0.5 around the estimate), a reference sample of 16 typically developing children from each age band (i.e., n=16 18-24 months corrected age; n=16 30-36 months corrected age) will be needed. In total, we propose to recruit 40 children aged 18-36 months.

An estimate of the standard deviation of 0.3 for the typically developing group was based on a previous sample of typically developing children participating in the GNPA study aged four years (scored on the DDS). It is expected that the variability in the younger age range will be greater than the four year old sample, therefore a standard deviation of 0.5 was used to ensure the sample is large enough to give precision to the estimate of mean scores. The DDS is the

measure expected to have the greatest variability in scores, therefore it has been used for the sample size calculations.

Statistical considerations

This study explores the relationship between OPD as an outcome variable (overall, impairment in saliva control, oral and pharyngeal phases, food/ fluid textures) with the primary exposure variable of gross motor skill attainment. It also investigates OPD as an exposure variable for the outcomes of growth and nutritional status. The statistical analysis plan is summarised in Table 2. Demographic data of the sample will be presented with descriptive statistics, and sample representativeness to the population determined by comparing the prevalence of GMFCS classifications to the non-participants and data reported in an Australian register study[68].

Inter-rater and intra-rater reliability of the primary measures (SOMA, DDS, PSAS, pharyngeal signs, saliva control and GMFCS) will be assessed using Cohen's Kappas (weighted and unweighted), and percentage agreement will be used. Existing cut scores for the SOMA, DDS and PSAS will be evaluated for their sensitivity and specificity to accurately identify typically developing children as no oropharyngeal dysphagia. The mean score of the typically developing reference sample ($Mean_{TDC}$) + two standard deviations (SD) will be used to determine more appropriate cut scores for the measures (i.e., scores above 2SD of the $Mean_{TDC}$ are considered to indicate presence of oropharyngeal dysphagia). The reference sample will be included in regression analyses for the overall study as a base group for comparison.

Table 2 Summary of Primary Outcome and Exposure Variables in the Present Study by Objective, and Statistical Tests

Hypothesis	Outcome Variable	Exposure Variable	Statistics
H2(i)	**OPD overall (yes on SOMA, DDS, PSAS or clinical pharyngeal signs)** *Dichotomous*	**GMFCS** GMFM-88 domains MACs Motor type/ distribution	Prevalence, Chi square Binomial logistic regression
H2(i)	**SOMA (overall)** *Dichotomous* **DDS (overall)** *Dichotomous* **PSAS (overall)** *Dichotomous* **Pharyngeal signs (overall)** *Dichotomous* **Saliva control (overall)** *Dichotomous*	**GMFCS** GMFM-88 domains MACs Motor type/ distribution	Prevalence, Chi square Binomial logistic regression
H2(i)	**DDS Part 2 raw score** *Continuous* **Dysphagia Severity Score** *Ordinal*	GMFM-66 GMFCS	Linear regression Multinomial logistic regression
H2(ii)	**Growth (height/ length, knee and upper arm length) Ax1** *Continuous*	**OPD & subtypes Ax1**	Linear regression
H2(ii)	**Nutritional Status (skin-folds, BMI) Ax1** *Continuous*	**OPD & subtypes Ax1**	Linear regression
H3	**OPD, SOMA, DDS, Pharyngeal Signs, Saliva Control, Parent Report Ax2** *Dichotomous*	**OPD, SOMA, DDS, PSAS, Pharyngeal Signs, Saliva Control, Parent Report Ax1** *Dichotomous*	Chi square to compare prevalence Binomial logistic regression
H3	**OPD at Ax1** *Dichotomous*	**GMFCS (collapsed)**	Binomial logistic regression
H3	**OPD at Ax2** *Dichotomous*	**GMFCS (collapsed)**	Binomial logistic regression
H3	**Nutritional Interventions (Tube feeding and/ or supplements) Ax2** *Ordinal*	**OPD & subtypes Ax1**	Multinomial logistic regression
H3	**Growth (height/ length, knee and upper arm length) Ax2** *Continuous*	**OPD & subtypes Ax1**	Linear regression
H3	**Nutritional Status (skin-folds, BMI) Ax2** *Continuous*	**OPD & subtypes Ax1**	Linear regression

Key: Ax1 18-24 month assessment; Ax2 30-36 month assessment; BMI Body Mass Index; DDS Dysphagia Disorders Survey; GMFCS Gross Motor Function Classification System; GMFM Gross Motor Function Measure; MACs Manual Ability Classification System; OPD Oropharyngeal dysphagia; PSAS Pre-Speech Assessment Scale; SOMA Schedule for Oral Motor Assessment

The strength of relationship between outcome and exposure variables will be analysed using regression modelling with odds ratios (for binary outcome variables) and relative risk ratios (for ordinal outcome variables). Ninety-five percent confidence intervals will be calculated for all effect estimates. GMFCS levels will be collapsed into three groups (GMFCS I-II, GMFCS III and GMFCS IV-V) for regression models in the longitudinal study (n=60) to increase statistical power. All demographic data, such as age, gender and geographical location, will be used in regression models to explore potential confounding with the primary variables. Postcode will be used to allocate children into five geographical categories from highly accessible to very remote[69]. Likelihood ratios will be used to evaluate the influence of covariates on the models, using backward stepwise elimination. If a group within a model has perfect prediction of the outcome, odds ratios will be calculated after applying a continuity correction of 0.5 to each appropriate cell. All data analyses will be performed using Stata Statistical Software[70]. For all tests, significance will be set at $p<0.05$.

ETHICS AND DISSEMINATION

Ethics committee approvals have been gained through the University of Queensland Medical Research Ethics Committee (2008002260), the Children's Health Services District Ethics Committee (HREC/08/QRCH/112), the Mater Health Services Human Research Ethics Committee (1520EC), the Cerebral Palsy League of Queensland (CPLQ 2008/ 2010 1029), Gold Coast Health Service District Human Research Ethics Committee (HREC/09/QGC/88), Central Queensland Health Services District Human Research Ethics Committee (SSA/10/QCQ/13), and the Townsville Health Service District Human Research Ethics Committee (HREC/09/QTHS/96). There are no known health or safety risks associated with participation in any aspect of the described study. All families will give written informed consent to participate, and they are able to withdraw their child from the study at any time

without explanation, without any penalty from staff at the Royal Children's Hospital or University of Queensland, or any effect on their child's care. Data collected in this study will be stored in a coded re-identifiable form (by ID number). Each child has three assessment appointments across the duration of the study, which necessitates data to be re-identifiable.

To our knowledge, this protocol outlines the first large population-based study using direct clinical feeding assessment in young children with cerebral palsy. The results of this study are planned to be published in peer reviewed medical and clinical journals, and presented at relevant international conferences. The following publications are proposed:

- Validity and reproducibility of measures of orophayrngeal dysphagia for young children with cerebral palsy
- Oropharyngeal dysphagia in young children with cerebral palsy and its relationship to gross motor skills
- Oral phase impairment in young children with cerebral palsy
- Pharyngeal phase impairment in young children with cerebral palsy
- Functional feeding skills, food and fluid texture inclusion in diets of young children with cerebral palsy
- Longitudinal relationships between orophayrngeal dysphagia, gross motor skills, growth and nutritional status in young children with cerebral palsy

LIST OF ABBREVIATIONS

CP	Cerebral Palsy
DDS	Dysphagia Disorders Survey
GMFCS	Gross Motor Function Classification System
GMFM	Gross Motor Function Measure
MACs	Manual Ability Classification System
NHMRC	National Health and Medical Research Council
OMCC	Oral Motor Challenge Category
OMD	Oral Motor Dysfunction
OPD	Oropharyngeal dysphagia
PSAS	Pre-Speech Assessment Scale
RCH	Royal Children's Hospital (Brisbane)
SOMA	Schedule for Oral Motor Assessment
UOS	Upper Oesophageal Sphincter
VFSS	Videofluoroscopic Swallow Study

AUTHORS' CONTRIBUTIONS

KAB, KW, KB and RB contributed to the study protocol. PD, RB, KB, RW and KW contributed to study concept, design and grant writing. All authors read, critically revised and approved the final manuscript.

COMPETING INTERESTS

The authors declare they have no competing interests.

FUNDING STATEMENT

This work was supported by the National Health and Medical Research Council (NHMRC) Postgraduate Medical and Dental Scholarship (1018264 – KB), NHMRC Career Development Fellowship (APP1037220– RB) and NHMRC Research Grants (569605 and 465128).

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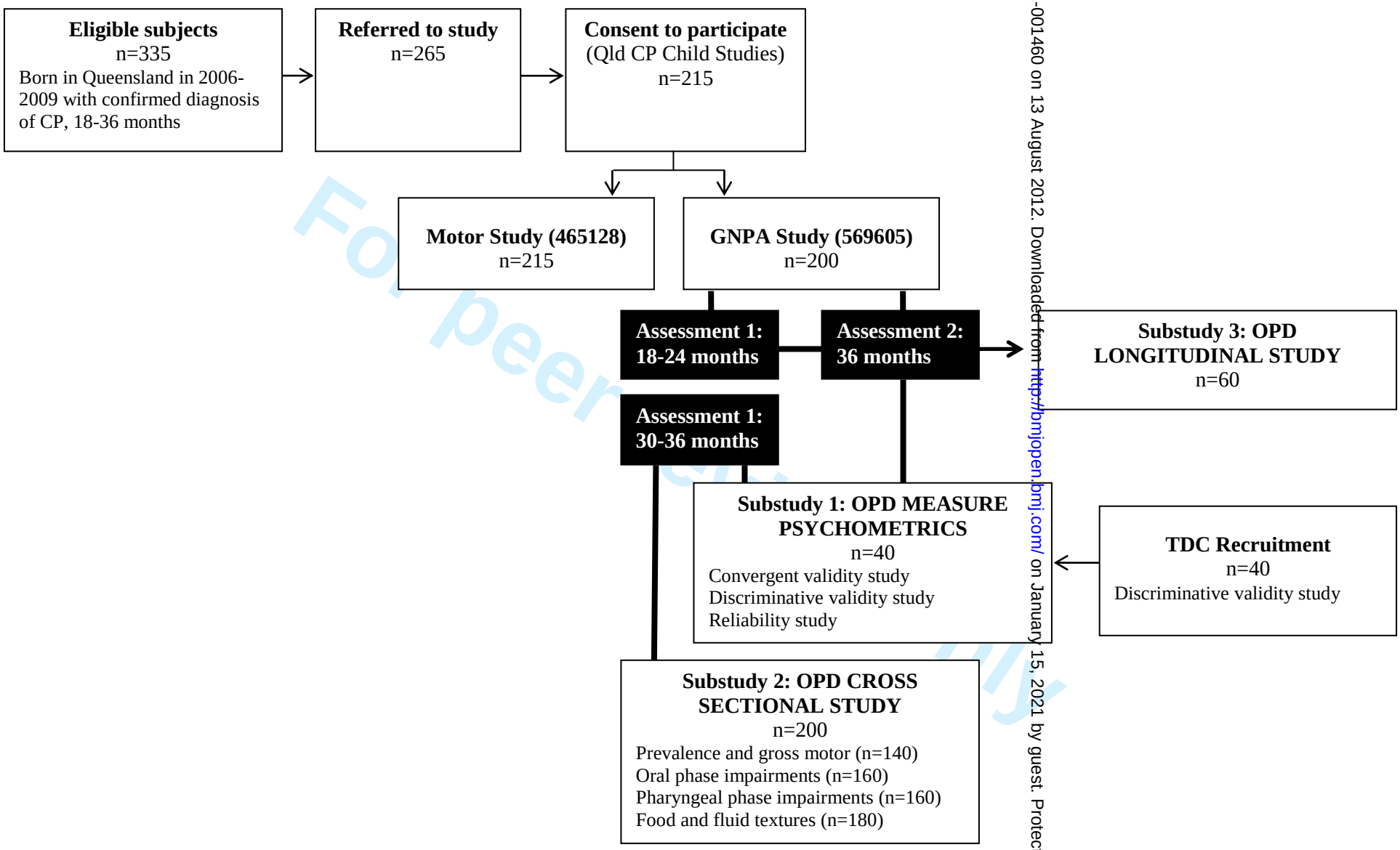

Correction

Benfer KA, Weir KA, Bell KL, *et al*. Longitudinal cohort protocol study of oropharyngeal dysphagia: relationships to gross motor attainment, growth and nutritional status in preschool children with cerebral palsy. *BMJ Open* 2012;**2**:e001460. A number of author corrections were inadvertently missed during the proofing stage:

1. The title of this paper should read: 'Protocol for a longitudinal cohort study of oropharyngeal dysphagia: relationships to gross motor attainment, growth and nutritional status in preschool children with cerebral palsy.'
2. Under the section "Aims and hypotheses" the expansion of DDS is actually "Dysphagia Disorders Survey" (not "Schedule").
3. Under the section "Thomas-Stonell & Greenburg Scale—saliva control", paragraph 2, reference 53 should be after "Scale" (as this is part of the measure name).
4. Table 2: H2(A) –These are all the same hypothesis, so all instances should have been in upper case A.

We apologise for these errors.

BMJ Open 2012;**2**:e001460corr1. doi:10.1136/bmjopen-2012-001460corr1