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RESEARCH PAPER

Oropharyngeal dysphagia in children with cerebral palsy: comparisons between a high- and low-resource country

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ABSTRACT

Purpose: There is paucity of research investigating oropharyngeal dysphagia (OPD) in young children with cerebral palsy (CP), and most studies explore OPD in high-resource countries. This study aimed at determining the proportion and severity of OPD in preschool children with CP in Bangladesh, compared to Australia.
Method: Cross-sectional, comparison of two cohorts. Two hundred and eleven children with CP aged 18–36 months, 81 in Bangladesh (mean = 27.6 months, 61.7% males), and 130 in Australia (mean = 27.4 months, 62.3% males). The Dysphagia Disorders Survey (DDS) – Part 2 was the primary OPD outcome for proportion and severity of OPD. Gross motor skills were classified using the Gross Motor Function Classification System (GMFCS), motor type/distribution.
Results: (i) Bangladesh sample: proportion OPD = 68.1%; severity = 10.4 SD = 7.9. Australia sample: proportion OPD = 55.7%; severity = 7.0 SD = 7.5. (ii) There were no differences in the proportion or severity of OPD between samples when stratified for GMFCS (OR = 2.4, $p = 0.051$ and $\beta = 1.2$, $p = 0.08$, respectively).
Conclusions: Despite overall differences in patterns of OPD between Bangladesh and Australia, proportion and severity of OPD (when adjusted for the functional gross motor severity of the samples) were equivalent. This provides support for the robust association between functional motor severity and OPD proportion/severity in children with CP, regardless of the resource context.

➤ IMPLICATIONS FOR REHABILITATION

- The proportion and severity of OPD according to gross motor function level were equivalent between high- and low-resource countries (LCs).
- Literature from high-resource countries may be usefully interpreted by rehabilitation professionals for low-resource contexts using the GMFCS as a framework.
- The GMFCS is a useful classification in LCs to improve earlier detection of children at risk of OPD and streamline management pathways for optimal nutritional outcomes.
- Rehabilitation professionals working in LCs are likely to have a caseload weighted towards GMFCS III–V, with less compensatory OPD management options available (such as non-oral nutrition through tubes).

Introduction

Oropharyngeal dysphagia (OPD) is common in up to 85% of children with cerebral palsy (CP), and is directly related to poor dietary intake and consequent undernutrition.[1,2] The feeding and swallowing process is commonly described in three distinct but overlapping phases which correspond to the anatomical structures involved in bolus transfer; oral-preparatory, oral (propulsive), and pharyngeal. OPD encompasses impairment to one or more phases of the swallow associated with eating, drinking or controlling saliva; and may include directly observed impairments (of the oral phase), or inferred impairments by observing clinical signs suggestive of impairment (as for the pharyngeal phase). With an estimated 80% of the global burden of CP in low-resource countries (LCs),[3] it is critical that greater research effort is focused on understanding functional outcomes of children with CP in these settings. The evidence-base available to health professionals working with children with OPD globally regarding OPD patterns and management strategies arises predominantly from high-resource countries. As such, an understanding of how these patterns of OPD compare to those in LCs will provide a foundation for clinicians working in LCs to interpret the existing OPD literature specific to their context. The use of the Gross Motor Function Classification System (GMFCS), a universally used classification in CP, may provide a robust framework to assist in this interpretation of outcome data from differing populations.

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ⓘ Supplemental data for this article can be accessed here

Amongst the limited literature on feeding and nutritional status in children with disabilities in LC, there is consensus that children's growth is compromised compared to non-disabled peers, which is consistent with literature from high-resource countries.[4–10] This poor growth has been linked to the child's inability to self-feed, however consideration of the influence of oral sensorimotor skills assessed using standardized measures has been minimal. One intervention study in Bangladesh investigated OPD in children with CP,[7] but neither the proportion of children with OPD at baseline, or analysis of OPD risk factors were reported. Three other studies have described the proportion of OPD in children with disabilities in LC, but have been limited to parent-report or informal methods.[4,11,12] Two studies (Turkey and India) estimated the prevalence of OPD in children with disabilities in LC at approximately 70%.[4,11] A study in South Africa found 35% of children with CP had been referred for feeding assessment based on retrospective chart review ($n = 19$), but did not discuss the proportion of confirmed OPD cases.[12]

This comparative study aims at exploring the proportion of children with OPD in Bangladesh, the location of the LC sample, in comparison to Australia, the location of the high-resource sample. Bangladesh is a densely populated country (approximately 150 million people residing in a country only 150,000 km^2),[13] where one third of people live in extreme poverty, and chronic malnutrition persists in 45% of children under five.[14] To our knowledge, only one population-based household survey in Bangladesh has been conducted, which estimated the prevalence of CP as 4/1000 live births.[15] In contrast to Bangladesh, Australia is a large sparsely populated country and considered a major global economy.[16] It was hypothesized that OPD in Bangladesh would be more frequent and severe compared to Australia when stratified for gross motor function due to delayed and decreased access to medical and rehabilitation services.

Methods

Study design and setting

This paper compares two cross-sectional studies of children with CP aged 18–36 months. The first sample is a population-based cohort of children born in Queensland, Australia, and the second a sample of clinic-attendees residing in Bangladesh. All children in the Australian sample accessed clinical care, so while recruitment methods differed between the samples, both were considered to represent children who would typically attend clinical services in their respective countries. The Australian data represents a subset of children from two larger longitudinal studies, Queensland CP Child Motor and Brain Development (National Health and Medical Research Council 465128) [17] and Queensland CP child: Growth, Nutrition and Physical Activity (National Health and Medical Research Council 569605).[18]

Participants

Children with a confirmed diagnosis of CP [19] aged 18–36 months corrected age were invited to participate in this study. Those with neurodegenerative conditions were excluded.

Additional recruitment criteria specific to the Australian sample included children born in Queensland from birth years 2006–2009; and only included initial assessments conducted between 1st January 2009 and 31st March 2013. Children of non-English speaking families were excluded due to lack of translation services.

The Bangladesh sample was recruited through in-patient services at a national tertiary rehabilitation facility, the Center for the Rehabilitation of the Paralyzed, from August to December 2013.

Variables and measures

Measures of the proportion and severity of OPD

Measures for the outcome of OPD are summarized in Table 1, with the Dysphagia Disorders Survey (DDS) selected as the primary outcome following comprehensive systematic review of the psychometric properties and clinical utility.[20,21]

The DDS – Part 2 consists of direct assessment of eight ingestion functions, assessing predominately the oral phase through a series of binary judgments (zero indicating no impairment, up to 22 indicating maximum impairment/non-oral nutrition). Primary validation and reliability were conducted in adults with developmental disability (mean 33 years), and shown to be strong.[22,23] The pediatric version has been used and validated in children from 18 months.[24,25] Users must be certified to rate the DDS, which has contributed to its excellent agreement within ratings (agreement 92.5%, kappa $= 0.4$ $p < 0.001$), and between raters (agreement $= 97.5\%$, kappa $= 0.7$ $p < 0.001$).[25] The smallest detectable change based on test-retest reproducibility between two mealtimes is 3.8 DDS points.[26] Modified cut-points were developed for children with CP aged 18–36 months to improve the specificity of the DDS when used in this young age range.[25] These cut-points were used in the present study to improve accurate detection of OPD.

An inference of pharyngeal phase dysphagia was noted if children demonstrated any one of 16 clinical signs, except a single cough on thin fluids.[27] Signs included gagging, coughing, choking, vomiting, throat clearing, multiple swallows, wheezing, stridor, rapid or labored breathing, wet breathing, gurgly voice, rattly chest, snuffly nose, eye tearing, or circumoral cyanosis/duskiness.[27] These signs have been validated in 150 children with dysphagia compared to videofluoroscopic swallow study (considered the gold standard for the pharyngeal phase). Wet voice (sensitivity 0.67, specificity 0.92, wet breathing (sensitivity 0.33, specificity 0.83) and cough (sensitivity 0.67, specificity 0.53) on thin fluids were considered good clinical markers.

The Thomas–Stonell and Greenberg saliva scale was used to rate presence and severity of saliva loss. It is a semi-quantitative, observational five-point ordinal scale (no loss to profuse).[28]

Table 1. Measures of oropharyngeal dysphagia.

	Direct assessment: Three components based on operational definition of OPD (any phase of swallow, during eating, drinking or saliva control)			Parent report
	Predominately oral phase in feeding	Pharyngeal phase (inferred) in feeding	Saliva loss	
Proportion with OPD (binary)	DDS	Sixteen clinical signs suggestive of pharyngeal phase impairment	Saliva loss	Parent report
Severity of OPD (continuous)	DDS part 2 raw score (0–22)	–	–	Visual analog scale for eating and drinking difficulties (0–10)

Key: Dysphagia Disorders Survey is the primary study outcome. DDS: Dysphagia Disorders Survey; OPD: oropharyngeal dysphagia.

Observation of saliva loss using standardized scales has been validated against weighed bibs (Spearman's $r = 0.604$, $p < 0.05$).[29]

Parents reported on their child's severity of eating problems and drinking problems on the CP Child Feeding Questionnaire using two 10 centimeter visual analog scales ("no problem" to "major problems").[26] The average of the two summed scales is used to indicate feeding severity based on parent report (with zero indicating no difficulties feeding and 10 indicating major difficulties). Presence of a feeding difficulty based on parent report was classified for severity scores greater than zero. The feeding questionnaire also gathered information on the presence of tube feeding.

Covariates and risk factors

Children's gross motor function was classified on the universally-adopted GMFCS using the <2 years and 2–4 year age-bands.[30] The type of CP (spasticity, dystonia, athetosis, hypotonia/ataxia) and motor distribution (unilateral versus bilateral) were also classified according to international guidelines.[31,32]

Gestational age (time between first day of the last menstrual period and the child's date of birth) was classified as term (>37 completed weeks of gestation), preterm, very preterm birth, and extremely preterm.[33]

The socio-economic status of Australian families was classified on the Socio-Economic Indexes for Areas Index of Relative Disadvantage.[34] In Bangladesh, the validated Poverty-Measurement Tool classified families into five levels from well-off to poor.[35]

Children's nutritional status was determined by height, weight, and body mass index. Height/length was measured using a portable stadiometer (Shorr Productions, MD, USA), and segmental lengths used when a direct measure of height was not possible. Weight was measured to the nearest 100 grams using digital scales. Anthropometric data were converted to z-scores according to World Health Organization reference data.[36]

Procedures

Children in Australia attended the hospital for diagnosis (based on published guidelines [19] and detailed clinical history gathered using the Physician Checklist [17]) and were followed-up for anthropometry, and videoed gross motor and mealtime assessments. In Bangladesh, children and their families attended the Center for the Rehabilitation of the Paralyzed for a two-week carer training and therapy program. On admission, they attended an appointment with the primary investigator (KB) and local Pediatric Consultant, who gathered a detailed clinical history using the Physician Checklist, and provided a preliminary diagnosis of CP. Throughout the two-week stay, children had mealtime and gross motor assessments videoed for later rating (by the Australian research team), and anthropometric measurements collected. Diagnosis was confirmed by a consistent Australian physician using the same diagnostic guidelines and data from the Physician Checklist, and supported by clinical presentation in the gross motor video. Two consistent Australian pediatric physiotherapists (experienced in CP) rated children's motor type/distribution and GMFCS.

All mealtime assessments were conducted according to a standardized snack protocol (described in greater detail in Benfer [20]) with children well positioned for three standardized presentations of four textures (puree, lumpy, chewable, and fluid), unless they were unsafe on a texture or refused. Following these standardized presentations, children were allowed to complete the snack as usual. Mealtime videos were later rated by the same pediatric speech pathologist (certified in the DDS) for both samples.

Ethics

All families gave written informed consent to participate. The Australian study was approved by the ethics committees of the Children's Health Services (Royal Children's Hospital Herston HREC 07/QRCH/107), Southern Health Ethics (05077C), University of Queensland (2007001784), Cerebral Palsy League of Queensland (CPLQ2008/2009 1010) and Mater Health Services (1186 C). Ethics for the Bangladesh Study were gained through the University of Queensland Medical Research Ethics Committee (2013000625), the Children's Health Services District Ethics Committee (HREC/13/QRCH/69), Center for the Rehabilitation of the Paralyzed Ethics Committee (CRP/RE/0401/55), and the International Center for Diarrhoeal Disease Research Bangladesh, Ethics Committee (PR-13047).

Data analyses

Characteristics of the Australian and Bangladesh samples were presented descriptively. Association between sample (Australia and Bangladesh) and sample characteristics were analyzed using logistic regression for binary outcomes; linear regression for continuous outcomes; and ordinal outcomes (GMFCS, motor type, preterm level) analyzed pairwise by level. Bangladesh was the comparison group, and models were adjusted for GMFCS, motor type, age, gender and preterm status (except when the covariate was the main explanatory variable). Association between sample and the OPD outcomes (proportion and severity) were analyzed using logistic (proportion) and linear (severity) regression, with GMFCS and sample as interaction terms to account for potential between country differences. Statistics demonstrating strength of relationship between variables (beta coefficient and odds ratio), and measures of fit (r squared and pseudo r squared) were reported. Univariate logistic regression analyzes were undertaken for all OPD risk factors of interest (age, gender, preterm status, epilepsy, GMFCS level, motor type and BMI z score). Variables consistently significant at the $p = 0.05$ level were then included in all multivariate regressions. Variance inflation factors less than 10 were considered to indicate no further testing for multicollinearity of covariates was required. All data analyzes were performed using Stata 10.0 (Statacorp 2007) with significance at $p < 0.05$.

Results

Differences in sample characteristics between countries

A total of 221 children with CP participated in this comparative study; 130 from Australia (AU) and 81 from Bangladesh (BD). For recruitment pathways see supplementary information 1. The mean age in months was equivalent between samples (AU: 27.4 SD 5.3, BD: 27.5 SD 6.1, $p = 0.80$). There was a significant association between sample and preterm status, motor type, GMFCS, and nutritional status (see Table 2).

Differences between countries in the proportion of children with OPD and OPD severity

The proportion of children with OPD based on direct assessment (on the DDS) in Bangladesh was 68.1% compared to 55.5% in Australia, however this did not differ significantly for any OPD subtype (see Table 2 for the proportion based on each of the OPD subtypes). Figure 1 shows the proportion of OPD by subtype,

Table 2. Characteristics of Australian and Bangladesh samples of preschool children with cerebral palsy.

Sample characteristic	Australia n (%)	Bangladesh n (%)	Crude OR (CI); p values (Bd base)	Adjusted OR (CI); p values
Gender				
Male	81 (62.3)	50 (61.7)	1.0 (0.6, 1.8); 0.93	1.0 (0.5, 1.8); 0.90
Female	49 (37.7)	31 (38.3)		
Preterm or term birth				
Extremely pre-term	19 (14.6)	0 (0.0)	19.3 (3.3, inf); <0.001[a]	NC[a]
Very pre-term	27 (20.8)	3 (3.9)	6.8 (2.0, 23.3); 0.002	7.3 (2.1, 25.9); 0.002
Preterm	20 (15.4)	15 (19.5)	0.8 (0.4, 1.7); 0.55	0.6 (0.3, 1.3); 0.18
Term	64 (49.2)	59 (76.6)	0.4 (0.2, 0.7); 0.001	0.4 (0.2, 0.8); 0.007
Motor type				
Spasticity	113 (86.9)	50 (61.7)	3.9 (2.0, 7.5); <0.001	2.8 (1.3, 5.8); 0.007
Unilateral	41 (31.5)	5 (6.2)	7.0 (2.6, 18.6); <0.001	2.6 (0.8, 8.5); 0.12
Bilateral (2 limbs)	30 (23.1)	21 (25.9)	0.9 (0.5, 1.6); 0.64	0.3 (0.1, 0.8); 0.009
Bilateral (3–4 limbs)	42 (32.3)	24 (29.6)	1.1 (0.6, 2.0); 0.77	2.9 (1.3, 6.4); 0.009
Dystonia	2 (1.5)	15 (18.5)	0.07 (0.02, 0.3); <0.001	0.1 (0.02, 0.5); 0.005
Athetosis	4 (6.2)	7 (8.6)	0.3 (0.1, 1.2); 0.09	0.6 (0.2, 2.6); 0.54
Ataxia/hypotonia	11 (8.4)	9 (11.1)	0.7 (0.3, 1.7); 0.40	0.7 (0.3, 2.0); 0.55
GMFCS				
I	57 (44.2)	7 (8.6)	8.3 (3.5, 19.3); <0.001	8.2 (3.5, 19.6); <0.001
II	15 (11.6)	12 (14.8)	0.8 (0.3, 1.7); 0.49	0.8 (0.3, 1.8); 0.53
III	23 (17.8)	25 (30.9)	0.5 (0.3, 0.9); 0.028	0.4 (0.2, 0.8); 0.008
IV	12 (9.3)	14 (17.3)	0.5 (0.2, 1.1); 0.09	0.6 (0.2, 1.3); 0.17
V	23 (17.7)	23 (28.4)	0.5 (0.3, 1.0); 0.05	0.6 (0.3, 1.2); 0.12
Poverty status	NA		NA	NA
Well off	–	25 (31.6)	–	–
Moderately well off	–	27 (34.2)	–	–
Not so well off	–	15 (19.0)	–	–
Poor	–	7 (8.9)	–	–
Very poor	–	5 (6.3)	–	–
Unknown	–	2 (2.5)	–	–
Socio-economic status	–	NA	NA	NA
Least disadvantaged	48 (37.2)	–	–	–
Middle tertile	40 (31.0)	–	–	–
Most disadvantaged	41 (31.8)	–	–	–
Tube fed	16 (12.3)	0 (0.0)	15.8 (2.6, inf); <0.001[a]	NC
Nutritional status				
HAZ: mean (SD)	−0.9 SD 1.4	−2.5 SD 1.4	$\beta = 1.7$ (<0.001)	$\beta = 1.5$ (<0.001)
WAZ: mean (SD)	−0.3 SD 1.2	−2.4 SD 1.4	$\beta = 2.1$ (<0.001)	$\beta = 1.8$ (<0.001)
Underweight[b]	6 (4.6)	19 (23.5)	0.2 (0.1, 0.4); <0.001	0.3 (0.1, 0.8); 0.02
Proportion of OPD				
DDS	72 (55.5)	55 (68.1)	0.6 (0.3, 1.1); 0.09	4.1 (1.5, 11.4); 0.006
Clinical signs	81 (62.3)	49 (60.5)	1.0 (0.6, 1.7); 0.88	2.0 (1.0, 3.9); 0.06
Saliva loss	64 (49.2)	36 (44.4)	1.2 (0.7, 2.1); 0.51	2.3 (1.2, 4.7); 0.018
Parent report	78 (60.0)	57 (70.4)	0.6 (0.4, 1.1); 0.13	1.3 (0.6, 2.8); 0.438
Severity of OPD				
DDS mean (SD)	7.0 SD 7.5	10.4 SD 7.9	$\beta = -3.4$ (<0.001)	$\beta = 1.2$ (0.08)
Parent report mean (SD)	2.9 SD 3.5	3.6 SD 3.5	$\beta = -0.7$ (0.14)	$\beta = 0.8$ (0.05)

Adjusted odds ratio models include covariates of GMFCS, age, gender and preterm status, except when that variable is the main explanatory variable.
[a]Calculated using exact logistic regression as outcome predicts perfectly (adjusted OR not calculable).
[b]Based on BMI z score less than 2SD; Bd base Bangladesh comparison group.
CI: Confidence Interval; GMFCS: Gross Motor Function Classification System; HAZ: Height for age z score; inf: infinity; NA: Not applicable to the context, therefore odds ratios not calculated; NC: Not calculable as no children in Bangladesh in outcome; OR: Odds Ratio; WAZ: Weight for age z score.

stratified by GMFCS. Once stratified for GMFCS, the proportion of OPD was not significantly different between countries (OR = 2.4, $p = 0.051$, pseudo $r^2 = 0.31$), although this was significant for children classified as GMFCS I on the DDS, and GMFCS V on the clinical signs and parent report. The proportion of children with OPD (based on direct assessment on the DDS; clinical signs suggestive of pharyngeal phase impairment; observation of saliva loss; and parent-reported OPD) increased with poorer gross motor function for both samples, as presented in Figure 1); direct assessment on the DDS: AU: OR = 2.6 (95% CI = 1.8, 3.8), $p < 0.001$; BD: OR = 7.3 (95% CI = 2.8, 18.8), $p < 0.001$), direct observation of clinical signs AU: OR = 2.1 (95% CI = 1.6, 2.7), $p < 0.001$; BD: OR = 1.7 (95% CI = 1.2, 2.5), $p = 0.006$), direct assessment of saliva loss: AU: OR = 1.6 (95% CI = 1.2, 2.0), $p = 0.001$; BD: OR = 1.8 (95% CI = 1.2, 2.7), $p = 0.003$), parent-report: AU: OR = 1.7 (95% CI = 1.3, 2.3), $p < 0.001$; BD: OR = 2.6 (95% CI = 1.6, 4.2), $p < 0.001$).

OPD severity based on the DDS and parent-report, stratified by sample and GMFCS, is shown in Figure 2. Mean DDS score (SD) differed between samples (AU: 7.0 SD 7.5, BD: 10.4 SD 7.9; $\beta = -3.4$, $p < 0.001$, $r^2 = 0.05$), but was not significant after adjustment for differences in GMFCS distribution between samples ($\beta = 1.2$, $p = 0.08$, $r^2 = 0.66$). Children's gross motor function was significantly related to OPD severity for both samples (AU: $\beta = 3.8$, $p < 0.001$, $r^2 = 0.70$; BD: $\beta = 4.6$, $p < 0.001$, $r^2 = 0.56$). OPD severity based on parent-report was not significantly different between samples (AU: 2.9 SD 3.5, BD: 3.6 SD 3.5; $\beta = -0.7$, $p = 0.14$, $r^2 = 0.01$), even after adjusting for differences in GMFCS distribution ($\beta = 0.8$, $p = 0.051$, $r^2 = 0.43$). Children's OPD severity on specific textures (assessed on the DDS) differed between samples only for children in GMFCS V on non-chewables (mean score in BD 5.2 and 5.8 in AU, $p = 0.02$) and fluids (mean score in BD 4.8, and 5.7 in AU, $p = 0.03$).
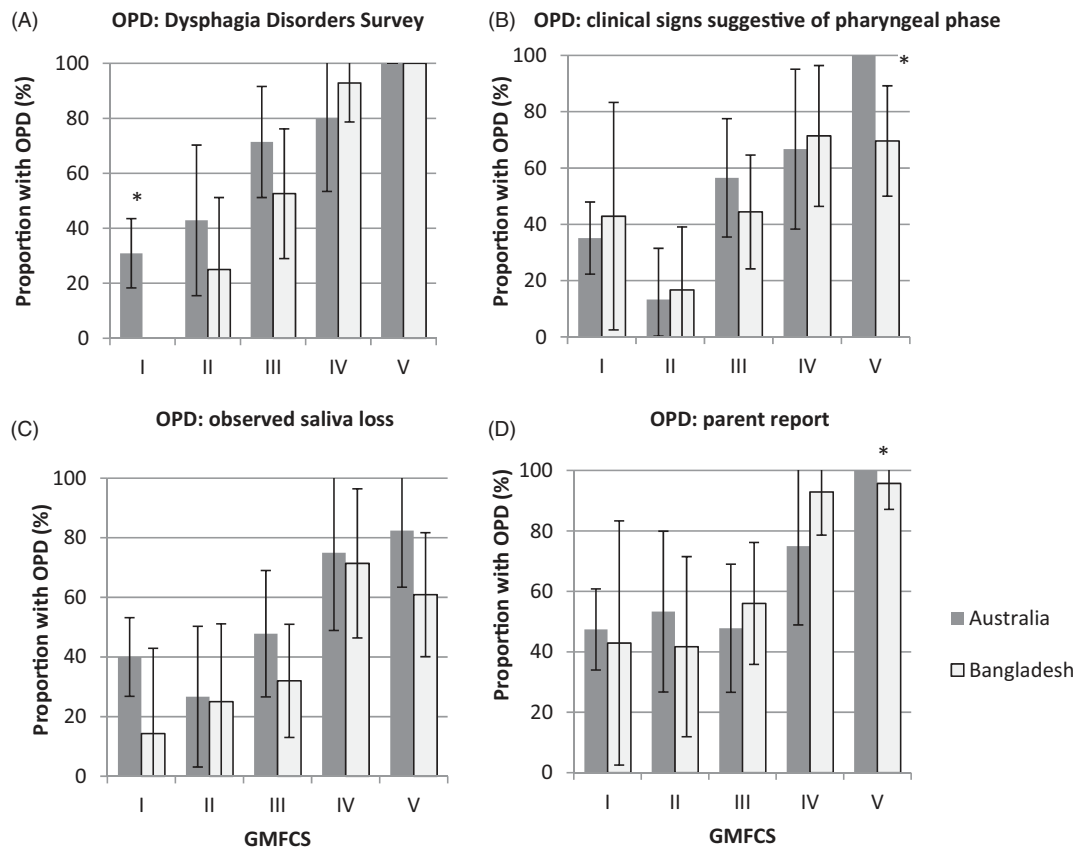
Figure 1. Key: *Indicates significant difference in proportion with OPD between samples; significantly more children in GMFCS I with OPD (DDS) in Australia compared to Bangladesh ($p < 0.001$); significantly more children in GMFCS V with clinical signs suggestive of pharyngeal phase impairment in Australia compared to Bangladesh ($p < 0.001$); significantly more children in GMFCS V with parent-reported OPD in Australia compared to Bangladesh; GMFCS: Gross Motor Function Classification System; OPD: oropharyngeal dysphagia.

Risk factors for OPD

The proportion and severity of OPD on the DDS was significantly associated with sample (i.e., country) in the multivariate models when adjusted for gross motor and demographic factors ($p < 0.001$, see Table 3). The odds of having OPD were 1.9–3.5 times greater with each increase in GMFCS. OPD severity was significantly greater with increasing GMFCS level. Preterm birth reduced children's likelihood of OPD based on the DDS, and resulted in lower OPD severity.

Discussion

Differences in sample characteristics between countries

The motor severity, motor type and other demographic characteristics of children with CP differed significantly between Australia and Bangladesh.[37] Of particular importance is the distribution of GMFCS levels and motor type, which are known to be related to patterns of OPD.[38] The Bangladesh sample was skewed towards children with poorer gross motor function (GMFCS III–V), whereas in Australia over half were classified as GMFCS I. In both samples, spasticity was the dominant motor type; however there were significantly fewer children with unilateral spasticity, and more children with dystonia in Bangladesh. This finding has implications for clinical services in Bangladesh, but may not be generalizable to the Bangladesh CP population, due to sampling of clinic attendees only.

Children with CP in Bangladesh had significantly lower height and weight z-scores, and more were classified "underweight"

compared to Australia, even after accounting for differences in GMFCS, preterm status, gender and age. OPD is one demonstrated risk factor for poor dietary intake and nutritional status in children with CP,[26,39] although there are also high background rates of malnutrition in children with typical development in Bangladesh.[14] More children in Australia used feeding tubes, which were absent in the Bangladesh sample and may also be a contributing factor to the rates of undernutrition in Bangladesh. While inherent differences in body size related to ethnicity exist, use of the WHO classification for 'underweight' status was considered to adequately account for these.

Differences between countries in the proportion of children with OPD and OPD severity

Overall, presence and severity of OPD was greater in the Bangladesh sample compared to Australia. Unadjusted OPD severity was on average 3.4 DDS units (out of 22) higher in the Bangladesh sample compared to Australian children, although within the margin of smallest detectable change for the DDS (based on previous reliability work conducted by our team using the DDS in a sample with equivalent characteristics).[26] The proportion of children with OPD in Bangladesh was comparable to estimates from other LCs.[4,11] Due to variability in the sample child characteristics and case ascertainment of previous research in LCs, comparisons are limited.

There was a trend for fewer children to have impaired saliva control in Bangladesh compared to Australia (although not significant). Children in Bangladesh had much lower fluid intakes, which

(A) **OPD severity on direct assessment (Dysphagia Disorders Survey)**
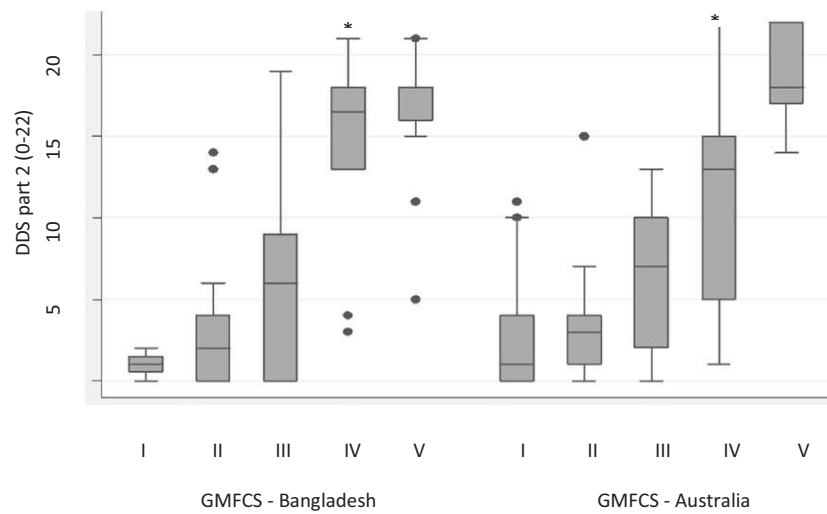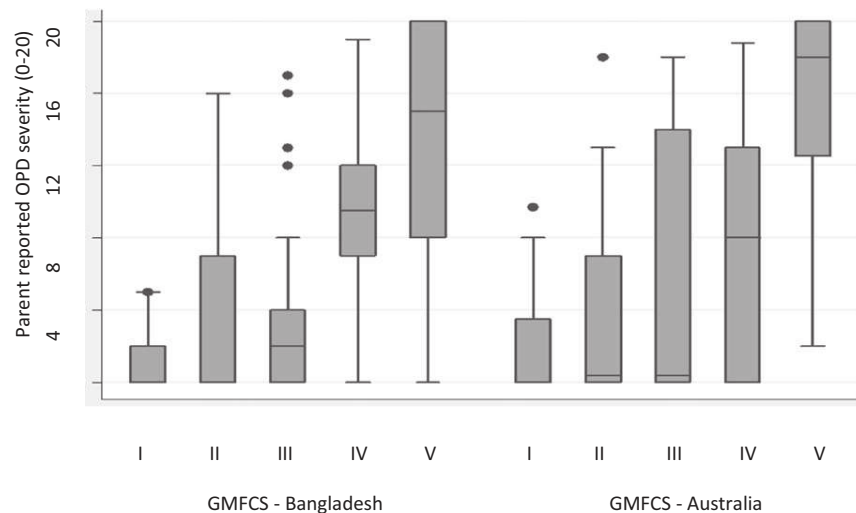

(B) **OPD severity on parent report**

Figure 2. Difference in mean DDS scores between samples by GMFCS level: I = 1.4 (−2.8, 5.5), p = 0.53; II = 0.4 (−2.8, 3.5), p = 0.83; III = 0.6 (−2.0, 3.2), p = 0.66, IV = −3.5 (−6.9, −0.2), p = 0.04; V = −2.3 (−0.1, 4.7), p = 0.060; Difference in mean parent-reported severity between samples by GMFCS level: I = 0.4 (−1.6, 2.5), p = 0.69, II = 0.0 (−2.0, 2.0), p = 1.0, III = −1.0 (−0.5, 2.4), p = 0.20; IV = −0.8, (−2.9, 1.3), p = 0.44; V = 1.2 (−0.3, 2.8), p = 0.10.

may have resulted in dehydration and lower saliva production. Anecdotally, mothers in Bangladesh frequently wipe children's saliva and food/fluid loss during the mealtime, so the observations of saliva loss pre/post mealtime (as per the snack protocol) may not accurately reflect children's saliva control in Bangladesh.

Relationship between OPD and GMFCS

Differences in the sample recruitment likely influenced the distribution of gross motor severity in these samples; as such, our *a priori* analysis plan was to stratify for GMFCS, accounting for some of these differences. Once stratified for GMFCS, differences in the proportion and severity of OPD between countries were minimal, with an overall trend for lower OPD proportions and severity in Bangladesh, contrary to our hypothesis. The only statistical differences were fewer children from GMFCS I having OPD (on the DDS) in Bangladesh, and fewer from GMFCS V having OPD (on the pharyngeal phase and parent report) in Bangladesh. This was

an unexpected finding, considering children in Bangladesh have later diagnoses, and later and less access to therapy.[37]

Children classified as GMFCS I from Australia had more varied scores, whereas in Bangladesh they were closely clustered around the mean. This subgroup was small in the Bangladesh sample, and was predominately children with bilateral spasticity with two-limb involvement. This differed markedly from the profile of children from GMFCS I in Australia with about 65% having unilateral spasticity. This may be a significant factor influencing the differences in OPD between countries. Furthermore, in Australia, children were more independent feeders, and encouraged to bring their own foods to the assessment from home (which corresponded to the standardized textures). In Bangladesh, children tended to be fed by their mothers, and were provided with the standardized foods at the assessment. These factors possibly increased the complexity of the mealtime in Australia, particularly for children with mild OPD.

A lower proportion of children from GMFCS V with clinical signs suggestive of pharyngeal dysphagia may be related to

Table 3. Risk factors for the presence and severity of oropharyngeal dysphagia in preschool children with cerebral palsy.

Sample characteristic	Adjusted statistic (CI); p values (Bd base)
DDS (overall), OR	
Sample	6.8 (1.9, 23.5); 0.003
GMFCS	**3.5 (2.2, 5.6); <0.001**
Motor type	**1.5 (1.0, 2.3); 0.037**
BMI	**0.9 (0.6, 1.4); 0.734**
Preterm	**0.2 (0.1, 0.6); 0.002**
Epilepsy	**1.8 (0.5, 6.1); 0.330**
DDS (severity), β	
Sample	**2.0 (0.4, 3.6); 0.017**
GMFCS	**3.8 (3.2, 4.3); <0.001**
Motor type	**0.6 (0.0, 1.1); 0.032**
BMI	**0.1 (−0.5, 0.6); 0.829**
Preterm	**−2.2 (−3.6, −0.8); 0.003**
Epilepsy	**0.4 (−1.2, 2.1); 0.603**
Pharyngeal phase, OR	
Sample	1.7 (0.8, 3.8); 0.184
GMFCS	**1.9 (1.4, 2.5); <0.001**
Motor type	1.0 (0.9, 1.3); 0.857
BMI	1.1 (0.9, 1.4); 0.343
Preterm	0.9 (0.4, 1.7); 0.628
Epilepsy	**1.1 (0.5, 2.5); 0.782**

Key: Bold indicates covariates which were significant <0.05 on univariate analysis for each outcome.
BMI: Body Mass Index; CI: Confidence Interval; DDS: Dysphagia Disorders Survey; GMFCS: Gross Motor Function Classification System; OR: Odds Ratio.

intrinsic and extrinsic factors. A much greater proportion of children from GMFCS V had dystonia in Bangladesh (30%) compared to only 4% in Australia. Also, mothers of children with non-ambulatory CP in Bangladesh, were more likely to deliver very small amounts of fluids to their child and in a controlled mode of delivery (often from a teaspoon), which may have had an influence on the presence of clinical signs.

While GMFCS-adjusted rates of OPD were lower in Bangladesh, the nutritional status of children told a different story. Specific exploration of the association between OPD and nutritional status was beyond the scope of the current analysis. It is well documented that OPD is only one risk factor of poor nutritional status, and as such the influence of other risk factors of poor nutritional status universally present in the pediatric population in Bangladesh warrants more detailed analysis. The series of binary judgments which constitute to the DDS raw score (used to indicate OPD severity) do not account for graded differences within each ingestion function. Subjectively, it seemed that children in Bangladesh had more significant OPD, but that the DDS may have lacked sensitivity to detect such differences. Exploring other markers of OPD, such as safety on food/fluid textures, mealtime duration, habitual dietary and texture intake, in addition to the standardized clinical assessment on the DDS, may provide a more comprehensive picture of the child's OPD.

Risk factors

The prevalence and severity of OPD increased markedly with each increase in GMFCS for Australia and Bangladesh. This was consistent with previous literature on this relationship.[24,38,40–44] Even after adjusting for other important health and demographic risk factors, GMFCS remained strongly related to OPD, suggesting that it is a helpful framework to be used in interpreting functional outcomes in varied samples such as this study. Preterm status also reduced the likelihood of OPD and resulted in lower severity on the DDS, independent of its association with motor type/severity.

This supports reports in the literature of poorer gross motor functional outcomes associated with later neurological lesions.[3,45]

Study limitations

While the strengths of this study included consistent raters and methods between countries, there were a number of limitations which may influence the interpretation of the findings. Most significant was the recruitment of clinic-attendees in Bangladesh, as opposed to a population-based sample in Australia. This limits generalizability to the population; although analysis of the findings by GMFCS allowed comparisons to be drawn between country samples. Furthermore, we anticipate that the analysis by gross motor functional severity presented in this study could be used in conjunction with population-based household surveys or register studies in LCs in order to estimate the population prevalence of OPD.

Due to recruitment of clinic-attendees in Bangladesh, only a small number of participants were classified as GMFCS I, as parents of children with minimal limitation to gross motor function appeared to be less likely to access services. Consequently wide confidence intervals were obtained for this sub-group in the OPD prevalence estimates, which should be interpreted with caution. The homogeneity of OPD severity in children from Bangladesh classified as GMFCS I, however, meant that the small numbers had minimal impact on this analysis.

Conclusions

Despite overall qualitative differences in the specific limitations to ingestion functions and mealtime behaviors between the Bangladesh and Australian samples, the proportion and severity (when adjusted for the functional gross motor severity of the samples) were equivalent. Children participating in the Bangladesh sample represent those commonly accessing services in Bangladesh, and as such mealtime and nutritional strategies to support optimal health for children with non-ambulatory CP need greater focus and implementation. This study also provides support for the robust association between functional gross motor severity and OPD proportion/severity in young children with CP, regardless of the resource context. We propose this may be a useful framework to apply when interpreting CP outcome literature arising from varied populations globally, which is particularly important for clinicians working in contexts with limited health research. In addition, application of the GMFCS as an early classification system administered by the multidisciplinary team may contribute to improved screening and earlier intervention of children with CP and OPD in LCs.

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Disclosure statement

The authors declare they have no conflict of interest.

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References

[1] Stevenson RD, Haves RP, Cater LV, et al. Clinical correlates of linear growth in children with cerebral palsy. Dev Med Child Neurol. 1994;36:135–142.

[2] Krick J, Van Duyn MS. The relationship between oral-motor involvement and growth: a pilot study in a pediatric population with cerebral palsy. J Am Diet Assoc. 1984;84:555–559.

[3] Gladstone M. A review of the incidence and prevalence, types and etiology of childhood cerebral palsy in resource-poor settings. Ann Trop Pediatr. 2010;30:181–196.

[4] Yousafzai AK, Filteau S, Wirz S. Feeding difficulties in disabled children leads to malnutrition: experience in an Indian slum. Br J Nutr. 2003;90:1097–1106.

[5] Tompsett J, Yousafzai AK, Filteau SM. The nutritional status of disabled children in Nigeria: a cross-sectional survey. Eur J Clin Nutr. 1999;53:915–919.

[6] Socrates C, Grantham-McGregor SM, Harknett SAJ. Poor nutrition is a serious problem in children with cerebral palsy in Palawan, the Philippines. SA J Trop Med Public Health. 1997;28:50–68.

[7] Adams MS, Khan NZ, Begum SA, et al. Feeding difficulties in children with cerebral palsy: low-cost caregiver training in Dhaka, Bangladesh. Child Care Health Dev. 2012;38:878–888.

[8] Pai M, Alur M, Wirz S, et al. A pilot study of the nutritional status of disabled and non-disabled children living in Dharavi, Mumbai. J Pediatr. 2001;38:60–65.

[9] Ojofeitimi EO. Nutritional status of handicapped pupils in Ile-Ife, Nigeria. Trop Doct. 1983;13:181–183.

[10] Alakija W. Nutritional status of normal and handicapped children in Bennin City, Nigeria. Trop Doct. 1988;18:33–35.

[11] Sen E, Yurtsever S. Difficulties experienced by families with disabled children. J Spec Pediatr Nurs. 2007;12:238–252.

[12] Barratt J, Ogle V. Recorded incidence and management of dysphagia in an outpatient pediatric neurodevelopmental clinic. SA J Child Health. 2010;4:38–41.

[13] World Bank [Internet]. Data. 2014; [cited 2014 Dec 20]. Available from: http://data.worldbank.org/

[14] Rayhan MI, Khan MSH. Factors causing malnutrition in children under 5 in Bangladesh. Pak J Nutr. 2006;5:558–562.

[15] Bangladesh Ministry of Health and Family Welfare. Survey of autism and neurodevelopmental disorders in Bangladesh. Bangladesh: Sudipta Printers & Packagers Ltd.; 2013.

[16] G20 [Internet] G20 members. 2014; [cited May 16]; Available from: https://www.g20.org/about_g20/g20_members.

[17] Boyd RN, Jordan R, Pareezer L, et al. Australian Cerebral Palsy Child Study: protocol of a prospective population based study of motor and brain development of preschool aged children with cerebral palsy. BMC Neurol. 2013;13:57e57–57e69.

[18] Bell KL, Boyd RN, Tweedy SM, et al. A prospective, longitudinal study of growth, nutrition and sedentary behavior in young children with cerebral palsy. BMC Public Health. 2010;10:e179–e191.

[19] Badawi N, Watson L, Petterson B, et al. What constitutes cerebral palsy? Dev Med Child Neurol. 1998;40:520–527.

[20] 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.

[21] Benfer KA, Weir KA, Boyd RN. Clinimetrics of measures of oropharyngeal dysphagia for preschool children with cerebral palsy and neurodevelopmental disabilities: a systematic review. Dev Med Child Neurol. 2012;54:784–795.

[22] Sheppard JJ. Dysphagia disorders survey and dysphagia management staging scale (adult and pediatric applications): user's manual: Australian edition. Ryde, NSW: The Center for Developmental Disability; 2003.

[23] Sheppard JJ, Hochman R, Baer C. The dysphagia disorder survey: validation of an assessment for swallowing and feeding function in developmental disability. Res Dev Disab. 2014;35:929–942.

[24] Calis EA, Veugelers R, Sheppard JJ, et al. Dysphagia in children with severe generalized cerebral palsy and intellectual disability. Dev Med Child Neurol. 2008;50:625–630.

[25] Benfer KA, Weir KA, Bell KL, et al. Validity and reproducibility of measures of oropharyngeal dysphagia in preschool children with cerebral palsy. Dev Med Child Neurol. 2014;57:358–365.

[26] Benfer KA, Weir KA, Bell KL, et al. Longitudinal study of oropharyngeal dysphagia in preschool children with cerebral Palsy. And relationship to health outcomes. Arch Phys Med Rehab. 2016;97:552–560.

[27] Benfer KA, Weir KA, Bell KL, et al. Clinical signs suggestive of pharyngeal dysphagia in preschool children with cerebral palsy. Res Dev Disab. 2015;38:192–201.

[28] Thomas-Stonell N, Greenberg J. Three treatment approaches and clinical factors in the reduction of drooling. Dysphagia. 1988;3:73–78.

[29] Senner JE, Logemann J, Zecker S, et al. Drooling, saliva production, and swallowing in cerebral palsy. Dev Med Child Neurol. 2004;46:801–806.

[30] Palisano R, Rosenbaum P, Walter RS, et al. Development and reliability of a system to classify gross motor function in children with cerebral palsy. Dev Med Child Neurol. 1997;39:214–223.

[31] Cans C. Surveillance of cerebral palsy in Europe: a collaboration of cerebral palsy surveys and registers. Dev Med Child Neurol. 2000;42:816–824.

[32] Sanger TD. Pathophysiology of pediatric movement disorders. J Child Neurol. 2003;18:S9–S24.

[33] World Health Organization [Internet]. Preterm birth: Fact sheet N°363. 2013; [cited May 29]. Available from: http://www.who.int/mediacentre/factsheets/fs363/en/.

[34] Australian Bureau of Statistics [Internet]. Census of population and housing: socio-economic indexes for areas (SEIFA), Australia. 2011; [cited June 25]. Available from: http://www.abs.gov.au/ausstats/abs@.nsf/mf/2033.0.55.001.

[35] Bhuiya A, Mahmood SS, Masud Rana AKM, et al. A multidimensional approach to measure poverty in rural Bangladesh. J Health Popul Nutr. 2007;25:134–145.

[36] World Health Organization, UNICEF. WHO child growth standards and the identification of severe acute malnutrition in infants and children: a joint statement by the World Health Organization and the United Nations Children's Fund. Geneva, Switzerland: WHO Press; 2009.

[37] Benfer KA, Jordan R, Bandaranayake S, et al. Motor severity in children with cerebral Palsy studied in a high-resource and low-resource country. Pediatrics. 2014;134:e1594–e1602.

[38] Benfer KA, Weir KA, Bell KL, et al. Oropharyngeal dysphagia and gross motor skills in children with cerebral palsy. Pediatrics. 2013;131:e1553–e1562.

[39] Benfer KA, Weir KA, Bell KL, et al. Food and fluid texture consumption in a population-based cohort of preschool children with cerebral palsy: relationship to dietary intake. Dev Med Child Neurol. 2015;57:1056–1063.

[40] Fung EB, Samson-Fang L, Stallings VA, et al. Feeding dysfunction is associated with poor growth and health status in children with cerebral palsy. J Am Diet Assoc. 2002;102: 361–373.

[41] Parkes J, Hill N, Platt MJ, et al. Oromotor dysfunction and communication impairments in children with cerebral palsy: a register study. Dev Med Child Neurol. 2010;52: 1113–1119.

[42] Reilly S, Skuse D, Poblete X. Prevalence of feeding problems and oral motor dysfunction in children with cerebral palsy: a community survey. J Pediatr. 1996;129:877–882.

[43] Sullivan PB, Lambert B, Rose M, et al. Prevalence and severity of feeding and nutritional problems in children with neurological impairment: Oxford Feeding Study. Dev Med Child Neurol. 2000;42:674–680.

[44] Waterman ET, Koltai PJ, Downey JC, et al. Swallowing disorders in a population of children with cerebral palsy. Int J Pediatr Otorhinolaryngol. 1992;24:63–71.

[45] Himpens E, Van den Broeck C, Oostra A, et al. Prevalence, type, distribution, and severity of cerebral palsy in relation to gestational age: a meta-analytic review. Dev Med Child Neurol. 2008;50:334–340.