Oropharyngeal Dysphagia and Gross Motor Skills in Children With Cerebral Palsy

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KEY WORDS
deglutition disorders, dysphagia, feeding, cerebral palsy, prevalence

ABBREVIATIONS
CP—cerebral palsy
DDS—Dysphagia Disorders Survey
GMFCS—Gross Motor Function Classification System
GMFM—Gross Motor Function Measure
MACS—Manual Ability Classification System
OPD—oropharyngeal dysphagia
SOMA—Schedule for Oral Motor Assessment

Ms Benfer and Ms Weir were responsible for acquisition of data; Ms Benfer was responsible for analysis and interpretation of data; Prof Boyd, Ms Benfer, and Ms Weir drafted the manuscript; Profs Davies, Boyd, Drs Bell, and Ware and Ms Weir were responsible for study design and grant writing; Ms Weir and Prof Boyd were responsible for study supervision, and all authors critically reviewed and approved the final manuscript. This trial has been registered with the ANZTR Register (Trial Registration Number: ACTRN12611000616976).

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WHAT'S KNOWN ON THIS SUBJECT: Oropharyngeal dysphagia (OPD) prevalence is 19-99%. OPD based on parent-report is associated with gross motor skills in children with cerebral palsy (CP), however this underestimates prevalence. Almost all children with severe CP have dysphagia; little is known about mild CP.

WHAT THIS STUDY ADDS: The prevalence of directly assessed OPD in preschool children with CP is 85% (70% in GMFCS I; 100% in GMFCS V). OPD was prevalent even in mild CP. Gross motor functional capacity is strongly related to dysphagia severity and prevalence.

abstract

OBJECTIVES: To determine the prevalence of oropharyngeal dysphagia (OPD) and its subtypes (oral phase, pharyngeal phase, saliva control), and their relationship to gross motor functional skills in preschool children with cerebral palsy (CP). It was hypothesized that OPD would be present across all gross motor severity levels, and children with more severe gross motor function would have increased prevalence and severity of OPD.

METHODS: Children with a confirmed diagnosis of CP, 18 to 36 months corrected age, born in Queensland between 2006 and 2009, participated. Children with neurodegenerative conditions were excluded. This was a cross-sectional population-based study. Children were assessed by using 2 direct OPD measures (Schedule for Oral Motor Assessment; Dysphagia Disorders Survey), and observations of signs suggestive of pharyngeal phase impairment and impaired saliva control. Gross motor skills were described by using the Gross Motor Function Measure, Gross Motor Function Classification System (GMFCS), Manual Ability Classification System, and motor type/distribution.

RESULTS: OPD was prevalent in 85% of children with CP, and there was a stepwise relationship between OPD and GMFCS level. There was a significant increase in odds of having OPD, or a subtype, for children who were nonambulant (GMFCS V) compared with those who were ambulant (GMFCS I) (odds ratio = 17.9, P = .036).

CONCLUSIONS: OPD was present across all levels of gross motor severity using direct assessments. This highlights the need for proactive screening of all young children with CP, even those with mild impairments, to improve growth and nutritional outcomes and respiratory health. Pediatrics 2013;131:e1553–e1562.
Oropharyngeal dysphagia (OPD) is reported to be prevalent in 19% to 99% of children with cerebral palsy (CP), and may lead to inadequate food/ fluid intake and reduced mealtime safety. It is associated with prolonged mealtimes, poor growth and nutritional status, and potential respiratory consequences, which are a major cause of premature mortality. This study defines OPD as impairment to any component of the oral-preparatory, oral (propulsive), and/or pharyngeal phases of the swallow, associated with eating, drinking, and/or controlling saliva. The neurologic lesion that affects an individual's oropharyngeal sensorimotor skills may also influence their gross motor skills, although the extent and severity may vary. An individual's gross motor skills may also influence the maintenance of a stable feeding posture, which can affect eating and swallowing by altering the position and alignment of the oropharyngeal structures and restricting their mobility.

There is generally agreement that OPD is positively associated with the severity of gross motor impairment. Assessment of OPD may be conducted directly, using clinical and/ or instrumental evaluation (such as videofluoroscopy), or indirectly through parent report or chart reviews. To date, studies tended to base the estimates of OPD prevalence on indirect measures, and have mostly focused on school-aged children and those with more severe gross motor impairments. This limits our understanding of the prevalence and nature of OPD and its relationship with gross motor skills, particularly in young children and including those with mild gross motor severities. An enhanced understanding of OPD in this subpopulation and its relationship with gross motor skills is important to facilitate early screening and identification of children at risk for poor growth, nutrition, and respiratory health. The aim of this study was to determine the prevalence of OPD and its subtypes (oral phase, pharyngeal phase, and saliva control) using direct clinical assessment of feeding with standardized measures of OPD, and to investigate the association between gross motor functional skills and OPD. It was hypothesized that OPD would be present across gross motor severity levels, and increase in prevalence and severity as gross motor severity increased.

**METHODS**

This is a cross-sectional population-based study of preschool-aged children with CP, conducted in Queensland, Australia, between April 2009 and August 2012. It is part of a longitudinal study exploring the relationship among growth, nutrition, and physical activity (Queensland CP Child: Growth, Nutrition and Physical Activity, National Health and Medical Research Council 56960). The design of the larger study and current study have been described elsewhere. Ethics approval was gained through the University of Queensland Medical Research Ethics Committee (20080002260), the Children’s Health Services District Ethics Committee (HREC/08/QRCH/112), and other regional and organizational ethics committees (see protocol papers for full list). All families gave written informed consent to participate.

**Patients**

Children with a confirmed diagnosis of CP, 18 to 36 months corrected age at the time of initial assessment, and born in Queensland between 2006 and 2009, were invited to participate in the study. Children with neurodegenerative conditions were excluded from the study.

**Measures**

Measures of OPD were selected after conducting a comprehensive systematic review of the psychometric properties and clinical utility. Included measures were the following:

1. Schedule for Oral Motor Assessment (SOMA): a discriminative measure that identifies oral motor dysfunction in children according to skills that are typically mastered from 8 to 24 months. The SOMA is predominantly a test of oral phase dysfunction; however, some items pertain to the pharyngeal phase. The assessment of feeding position (upright with/without back support, upright with trunk support, semi-sitting, and supine) was used as a covariate in models.

2. Dysphagia Disorders Survey–Pediatric (Part 2) (DDS): an evaluative measure for screening signs of oral, pharyngeal, and esophageal phase dysphagia in children and adults with a developmental disability. Part 2 provides a raw score that indicates an individual's functional eating competency (maximum impairment raw score of 22) and this subtest has been used previously as a measure of OPD.

3. Clinical signs suggestive of pharyngeal phase impairment: a determination of pharyngeal phase impairment was noted if the child demonstrated any 1 of 16 signs, rated live, and from video by the speech pathologist (see Appendix).

4. Thomas-Stonell Greenberg Saliva Severity Scale: a semiquantitative assessment of drooling severity (1- to 5-point scale of no drooling to profuse drooling) based on observations of anterior saliva loss.
Function Measure-88 (GMFM-88) for domain scores, and the Rasch-analyzed GMFM-66. From this, children were classified on the Gross Motor Function Classification System (GMFCS) according to their age by using the <2 years and 2- to 4-year scales. The Manual Ability Classification Scale (MACS) was used to classify children’s functional upper limb skills. The type of CP (spastic, dyskinetic, hypotonic/ataxic) and motor distribution (hemiplegia, diplegia, quadriplegia) were classified according to the Surveillance of CP in Europe. Children’s ability to sit on a mat and maintain head upright for 10 seconds, and sit on a bench for 10 seconds with feet supported were used to indicate head and trunk instability, respectively.

Procedures

Children attended the hospital for mealtime and gross motor assessment. Mealtimes were videoed as recommended in the SOMA administration manual, with children well positioned in their typical mealtime seating. Three standardized presentations of 4 textures (puree, lumpy, chewable, and fluid) were presented by the primary carer, using their regular utensils. Following these standard presentations, the child was allowed to complete the snack as usual. A trained researcher recorded 4 signs suggestive of pharyngeal phase impairment, and severity of drooling before and after the mealtime. All gross motor ratings were conducted by 2 trained physiotherapists.

Reproducibility Study

Twenty children (4 from each GMFCS level) were selected randomly by an independent researcher for analysis of intrarater and interrater reproducibility of all OPD measures and interrater reproducibility for MACS. The clinicians rating the videos were blinded to reliability case status. Intrarater reliability ratings were performed 2 weeks after initial ratings. Interrater reliability ratings were completed independently by 2 speech pathologists for the OPD measures (K.A.B., K.A.W.), and 2 physiotherapists for MACS.

Statistical Analysis

Demographic data were presented with descriptive statistics, and sample representativeness determined in relation to an Australian register study using χ² test for trend. Inter- and intrarater reproducibility were assessed by using Cohen’s κ (unweighted and weighted) and percentage agreement. The association between gross motor skill attainment and OPD were analyzed by using the χ² test for trend, and individual motor categories compared using logistic regression. Univariate
logistic regression analyses were undertaken for all explanatory variables of interest (age, gender, geographical accessibility, preterm status, postural instability, and supported feeding position). Variables consistently significant at the $P = .05$ level were then included in all multivariate regressions. All data analyses were performed by using Stata (Stata Corp, College Station, TX).

**RESULTS**

**Sample Characteristics**

There were 166 eligible children referred, of which 122 parents consented to participate in the Growth, Nutrition and Physical Activity study, and 120 children completed the requirements to participate in the OPD study. Of the children who declined participation, 18 participated in only the concurrent CP Child Motor Study,13,14 and 26 declined both studies (8 because of study burden, 12 because of family circumstances, 2 were non-English speaking, 3 resided interstate, and 1 died). Participants’ ages ranged from 17 to 37 months corrected age at the time of assessment (mean = 27.0 months, SD = 5.2). Partial or total tube feeding was present in 10.8% of the sample at the time of assessment. Characteristics of the sample are presented in Table 1.

**Reproducibility of Measures**

The results from the OPD reproducibility study are presented in Table 2. The inter- and intrarater agreement were >90% for all binary OPD measures, and reliability was substantial to perfect ($P < .001$). There was a strong correlation between DDS Part 2 scores for intrarater (intraclass correlation coefficient = 0.99, $P < .001$) and interrater (intraclass correlation coefficient = 0.99 $P < .001$). The MACS had 60% perfect agreement, and 36% near perfect agreement (within 1 level), with moderate reliability (weighted $\kappa = 0.47$, $P < .001$).

**Prevalence of OPD and Its Relationship With Motor Function**

Overall, 85% of children had OPD identified on 1 or more direct clinical measures (SOMA, DDS, or pharyngeal signs), excluding impaired saliva control, which was considered developmentally appropriate (Table 3). There was a significant increasing trend for the prevalence of all OPD variables as GMFCS level increased ($P < .05$) (Fig 1).

The relationships between GMFCS and OPD are presented in Table 3. Postural instability and position were consistently significantly associated with the OPD outcomes in all univariate models (largest $P$ value for instability was .008, and for position was .052). No other explanatory variables were significant for any outcome (with the exception of geographical access for pharyngeal signs, $P = .009$); therefore, instability and position were included in all

**Table 3** Relationship Between GMFCS and OPD

<table>
<thead>
<tr>
<th>GMFCS</th>
<th>Number (%)</th>
<th>Crude Odds Ratio (95% CI)</th>
<th>P Value</th>
<th>Adjusted Odds Ratio* (95% CI)</th>
<th>P Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>I</td>
<td>28 (56.0)</td>
<td>1.0 (reference)</td>
<td>—</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>II</td>
<td>14 (27.0)</td>
<td>2.0 (0.5–8.0)</td>
<td>.127</td>
<td>1.0 (reference)</td>
<td>—</td>
</tr>
<tr>
<td>III</td>
<td>11 (22.0)</td>
<td>1.0 (1.0–3.9)</td>
<td>.958</td>
<td>1.0 (reference)</td>
<td>—</td>
</tr>
<tr>
<td>IV</td>
<td>5 (10.0)</td>
<td>2.0 (0.5–8.0)</td>
<td>.127</td>
<td>1.0 (reference)</td>
<td>—</td>
</tr>
<tr>
<td>V</td>
<td>2 (4.0)</td>
<td>2.0 (0.5–8.0)</td>
<td>.127</td>
<td>1.0 (reference)</td>
<td>—</td>
</tr>
</tbody>
</table>

* Adjusted odds ratios for perfectly predicted are reported as $\approx 0.95$ CI n/c.

**Exposure predicts outcome perfectly, therefore calculated in eipsheet, based on Fisher’s Exact Test.**

CI, confidence interval; n/c, not calculable; —, no data available.

*CI on 1 or more direct clinical measures (SOMA, DDS, or pharyngeal signs), excluding impaired saliva control, which was considered developmentally appropriate (Table 3). There was a significant increasing trend for the prevalence of all OPD variables as GMFCS level increased ($P < .05$) (Fig 1).

The relationships between GMFCS and OPD are presented in Table 3. Postural instability and position were consistently significantly associated with the OPD outcomes in all univariate models (largest $P$ value for instability was .008, and for position was .052). No other explanatory variables were significant for any outcome (with the exception of geographical access for pharyngeal signs, $P = .009$); therefore, instability and position were included in all
multivariate models. For the overall OPD model, postural instability and supported feeding position perfectly predicted the presence of OPD (that is, no children with head or trunk instability, or fed in a supported feeding position had typical oral feeding skills); therefore, adjusted odds ratios could not be reported. The relationship between OPD and MACS is presented in Table 4. The results showed a similar finding to GMFCS models, with children classified in the severe levels for manual ability having significantly higher odds of OPD.

The relationship between OPD prevalence and gross motor capacity overall and by motor domain is reported in Table 5. For each unit increase in GMFM score, the odds of having OPD increased by 2% to 11% (odds ratio = 0.98 and 0.89, respectively). The severity of OPD, based on the DDS Part 2 raw score, was significantly correlated with motor severity on the GMFM-66 (Fig 2). The relationship between motor type/distribution and prevalence of OPD are presented in Table 6. All children with 4-limb involvement had OPD. Of the children with diplegia (spastic) and OPD, GMFM-66 scores were significantly lower than those with diplegia and no dysphagia (GMFM = 53.9, SD = 8.2 for OPD, compared with GMFM = 60.6, SD = 4.6 for no OPD (P = .028)). A similar but not statistically significant trend was noted in the unilateral spasticity group, of a GMFM score of 57.4, SD = 7.3, for OPD compared with 61.9, SD = 11.34, for those without OPD (P = .174).

DISCUSSION

This population-based study found 85% of children with CP aged 18 to 36 months had OPD, based on impairment on 1 or more of the SOMA, DDS, or pharyngeal signs. Estimates for the subtypes varied markedly from 43% on the SOMA, to 78% on the DDS, both of which are primarily measures of oral phase impairment. Pharyngeal phase impairment and impaired saliva control, identified through standardized clinical observations, were present in about half of the sample (62% and 48%, respectively).

The overall OPD estimate is consistent with the prevalence estimates reported in the only 2 previous studies conducted in preschool children with CP of 78%24 and 90%.11 However, the estimate by Reilly et al11 of 90%, obtained through direct assessments of a community-based sample, used the SOMA alone, which is a significantly higher estimate than ours of 40% found using only the SOMA. This discrepancy likely reflects the bias toward recruitment of participants with more severe gross motor impairments in the study by Reilly et al11 (70% had severe-profound motor impairment), when in fact the distribution of gross motor severity tends to be skewed to the milder end of the range.23 The estimate in the study by Wilson and Hustad24 was based on clinical evidence of oral-motor involvement, which was a broad classification of any neurologically based impairment of speech subsystems. Tube feeding or cough/choke/gag were identified by parent report in close to all of these children (73%). The gross motor severity of children in this study sample was not reported, thus their estimate may also reflect a bias toward recruitment of children with more severe gross motor severities. The current study estimate strengthens previous estimates by using direct clinical OPD measures with strong reproducibility and a representative study sample across gross motor severity levels.

There were an increasing number of children with OPD for each increase in GMFCS level, and this difference between groups was statistically significant for each subtype. OPD was present across all gross motor severity levels, with as few as 18% of children in GMFCS I identified as having OPD using the SOMA, and as many as 56% of children in this group using the DDS. Although the trend for increasing prevalence of OPD with increased gross motor severity levels.

![Proportion of oropharyngeal dysphagia by subtype, according to GMFCS. Key: OPD P < .001; SOMA P < .001; DDS P < .001; saliva control P = .002; pharyngeal signs P < .001.](attachment:FIGURE_1)
was stepwise for each GMFCS level, these relationships were generally only significant for children in GMFCS III to V compared with GMFCS I. All children who were tube fed were from GMFCS IV to V.

The proportion of children with OPD from the more severe gross motor groups was consistent with other studies. Direct ratings of children’s mealtimes were conducted in the studies by Calis et al.\(^2\) and Santoro et al.\(^25\) finding OPD in almost all children (99% and 100% respectively) from GMFCS IV to V, which was also found in the current study. In a large registry-based study of children (median age 5 years) \((n = 1357)\), there was a 5-fold increase in odds for GMFCS IV and a 15-fold increase for GMFCS V of having swallowing/chewing difficulties and excessive drooling.\(^1\) This increase in likelihood with GMFCS is comparable to the magnitude found in the current study, although the prevalence of OPD overall and by GMFCS level was markedly higher in the current study by using direct assessments. Using validated measures (SOMA and Standard Recording of Central Motor Deficit), 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.\(^11\) Although strengthened by using validated measures for both oral motor and gross motor skills, the sample was small, skewed to more severe gross motor severity levels, and only binary variables were used (presence/absence of dysfunction).

Children with mild gross motor impairments have received limited attention in the literature to date. A study of ambulatory (with or without assistive mobility) school-aged children (estimated to be GMFCS I to III) with mild CP performed more poorly than controls on spoon feeding, biting, and cup drinking using direct clinical assessment on the Functional Feeding Assessment modified.\(^26\) The specific prevalence of feeding difficulties or influence of gross motor skill could not be ascertained from the data. Another study investigating parent-reported feeding difficulties in children using GMFCS to classify gross motor level identified just 4% of children from GMFCS I to III with feeding difficulties, compared with 22% of children from GMFCS IV to V.\(^27\) The prevalence of OPD in the mild motor groups found in the current study are greater than previously documented, and may indicate the underdetection of mild feeding difficulties, particularly when using indirect assessments. The background prevalence in typically developing children and the potential effects of mild OPD on health warrant further investigation.

The results showed that the higher the gross motor capacity score on the GMFM, the fewer children had OPD (2% to 11% reduced chance of OPD with each increase in GMFM-66 score). This was statistically significant overall and for the 30- to 36-month stratum, but not for the 18- to 24-month group. Eighteen to 24 months is a period of significant gross motor maturation, which could explain the insignificant association in this age range. Almost three-quarters of the variability seen in the severity of OPD (by DDS part 2 raw score) could be explained by gross motor functional capacity. The gross motor domain with the greatest association with OPD

<table>
<thead>
<tr>
<th>OPD overall</th>
<th>I</th>
<th>II</th>
<th>III</th>
<th>IV</th>
<th>V</th>
<th>Odds Ratio (95% CI)</th>
<th>P value</th>
<th>Adjusted Odds Ratio* (95% CI)</th>
<th>P value</th>
</tr>
</thead>
<tbody>
<tr>
<td>n (%)</td>
<td>31 (70.5)</td>
<td>39 (88.9)</td>
<td>5 (100.0)</td>
<td>9 (100.0)</td>
<td>18 (100.0)</td>
<td>1.0 (reference)</td>
<td>3.5 (0.4–45.8)</td>
<td>0.013</td>
<td>33.5 (1.8–610.0)</td>
</tr>
</tbody>
</table>

**TABLE 4 Relationship Between Manual Ability and OPD**

*Confidence interval; n/c, not calculable.

* Multivariate models include postural instability and position. These covariates did not reach statistical significance for any outcome. Odds ratios for perfectly predicted are reported as \(\infty\) (95% CI n/c).

* Exposure predicts outcome perfectly, therefore calculated in epischet, based on Fisher’s Exact Test.

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Chi-square tests of independence were used to determine the significance of the relationship between OPD overall and by GMFCS level. The prevalence of OPD was significantly associated with the presence of gross motor impairment in children using specific GMFCS levels, with higher GMFCS levels associated with a higher prevalence of OPD. The association was stepwise for each GMFCS level, with the greatest association observed for GMFCS V. The adjusted odds ratios for OPD overall and by GMFCS level were calculated using multivariate models that included postural instability and position. These covariates did not reach statistical significance, but their inclusion improved the model fit. The adjusted odds ratios ranged from 3.5 (GMFCS II) to 45.8 (GMFCS IV) for OPD overall, and 3.3 to 610.0 for OPD by GMFCS level. The adjusted odds ratios for OPD overall and by GMFCS level were calculated using multivariate models that included postural instability and position. These covariates did not reach statistical significance, but their inclusion improved the model fit. The adjusted odds ratios ranged from 3.5 (GMFCS II) to 45.8 (GMFCS IV) for OPD overall, and 3.3 to 610.0 for OPD by GMFCS level.
prevalence was the sitting domain, which is consistent with the literature, that suggests postural stability and trunk control are important for feeding success.2,8 This was further supported by the influence of postural instability (head and trunk) and feeding position on the GMFCS model.

The number of children with OPD was strongly linked to motor type and distribution, with distributions affecting 3 or more limbs (spastic quadriplegia, dyskinesias, and hypotonia/ataxia) resulting consistently in OPD. As dyskinesias and hypotonia are less common in the CP population, the small numbers meant that these relationships were not statistically significant, despite OPD being consistently present.

This finding is consistent with the Oxford Feeding Studya of 271 school-aged children with OPD, which found that those with more extensive motor involvement (ie, quadriplegia and dyskinesia) are most likely to have difficulties with swallowing and articulation. In our study, about 70% of children with hemiplegia or diplegia had OPD, and these children had lower average scores on the GMFM than children with hemiplegia/diplegia and no feeding difficulties. This suggests that motor type/distribution in conjunction with functional severity may be useful in predicting children at risk for feeding difficulties.

This study explored OPD in a large representative sample of young children with CP using direct clinical measures. Although overall the sample size was adequate, the lower occurrence of certain phenomena in children with CP limited the statistical power for some individual analyses. Another limitation in this study was sampling an age range that crossed 2 different GMFCS scales (<2 years and 2–4 years). Children’s GMFCS level may be reclassified after their second birthday, which may affect comparisons across the sample. The most significant limitation in all studies of feeding in young children remains the lack of a gold standard or consensus in the definition for the construct of OPD. A large range was found in the identification of OPD cases using each of the OPD measures, particularly between the SOMA and DDS. The SOMA was designed to detect clinically significant OPD, and therefore may lack sensitivity in detecting mild OPD. Conversely, although the DDS and pharyngeal signs appear to be detecting the milder feeding difficulties, these measures may be misclassifying behaviors as OPD that are present in young typically developing children. Although normative data exist for feeding efficiency28,29 and parent-reported acquisition of a limited number of oral behaviors,30 our future work assessing a typically developing reference sample with the SOMA and DDS will address some of the questions surrounding the validity of measures. Triangulation of videofluoroscopy swallow study results with clinical pharyngeal signs will further validate these findings, and will be the subject of future articles.

**CONCLUSIONS**

This study proposes a more plausible OPD estimate of 85% to reflect the prevalence of OPD in young children with CP based on direct clinical measures with reported validity and reliability, and a representative population-based sample across the range of gross motor functioning.

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**TABLE 5** Relationship Between Motor Capacity on the GMFM and Prevalence of OPD

<table>
<thead>
<tr>
<th>OPD</th>
<th>Odds Ratio</th>
<th>95% Confidence Interval</th>
<th>P Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>GMFM-66 overallb</td>
<td>0.89</td>
<td>0.84–0.95</td>
<td>.001</td>
</tr>
<tr>
<td>18–24 mo</td>
<td>0.90</td>
<td>0.81–1.00</td>
<td>.054</td>
</tr>
<tr>
<td>30–36 moa</td>
<td>0.93</td>
<td>0.88–0.99</td>
<td>.017</td>
</tr>
<tr>
<td>GMFM-88 (A) lying, rollingb</td>
<td>0.96</td>
<td>0.92–0.99</td>
<td>.020</td>
</tr>
<tr>
<td>18–24 mo</td>
<td>0.95</td>
<td>0.89–1.01</td>
<td>.126</td>
</tr>
<tr>
<td>30–36 moa</td>
<td>0.97</td>
<td>0.93–1.01</td>
<td>.121</td>
</tr>
<tr>
<td>GMFM-88 (B) sittingb</td>
<td>0.92</td>
<td>0.87–0.97</td>
<td>.004</td>
</tr>
<tr>
<td>18–24 mo</td>
<td>0.95</td>
<td>0.88–1.01</td>
<td>.078</td>
</tr>
<tr>
<td>30–36 moa</td>
<td>0.90</td>
<td>0.82–0.98</td>
<td>.017</td>
</tr>
<tr>
<td>GMFM-88 (C) crawling, kneelingb</td>
<td>0.96</td>
<td>0.94–0.99</td>
<td>.002</td>
</tr>
<tr>
<td>18–24 mo</td>
<td>0.98</td>
<td>0.95–1.01</td>
<td>.118</td>
</tr>
<tr>
<td>30–36 moa</td>
<td>0.95</td>
<td>0.91–0.99</td>
<td>.015</td>
</tr>
<tr>
<td>GMFM-88 (D) standingb</td>
<td>0.96</td>
<td>0.94–0.98</td>
<td>.001</td>
</tr>
<tr>
<td>18–24 mo</td>
<td>0.97</td>
<td>0.94–1.00</td>
<td>.050</td>
</tr>
<tr>
<td>30–36 moa</td>
<td>0.95</td>
<td>0.92–0.99</td>
<td>.013</td>
</tr>
<tr>
<td>GMFM-88 (E) walking, running, jumpingb</td>
<td>0.96</td>
<td>0.94–0.98</td>
<td>.000</td>
</tr>
<tr>
<td>18–24 mo</td>
<td>0.95</td>
<td>0.92–1.00</td>
<td>.027</td>
</tr>
<tr>
<td>30–36 moa</td>
<td>0.96</td>
<td>0.93–0.99</td>
<td>.007</td>
</tr>
</tbody>
</table>

a Crude odds ratios reported as covariates of postural instability and position predict perfectly for outcome.
b Statistically significant.

despite OPD being consistently present. The number of children with OPD was strongly linked to motor type and distribution, with distributions affecting 3 or more limbs (spastic quadriplegia, dyskinesias, and hypotonia/ataxia) resulting consistently in OPD. As dyskinesias and hypotonia are less common in the CP population, the small numbers meant that these relationships were not statistically significant, despite OPD being consistently present. This finding is consistent with the Oxford Feeding Studya of 271 school-aged children with OPD, which found that those with more extensive motor involvement (ie, quadriplegia and dyskinesia) are most likely to have difficulties with swallowing and articulation. In our study, about 70% of children with hemiplegia or diplegia had OPD, and these children had lower average scores on the GMFM than children with hemiplegia/diplegia and no feeding difficulties. This suggests that motor type/distribution in conjunction with functional severity may be useful in predicting children at risk for feeding difficulties.

This study explored OPD in a large representative sample of young children with CP using direct clinical measures. Although overall the sample size was adequate, the lower occurrence of certain phenomena in children with CP limited the statistical power for some individual analyses. Another limitation in this study was sampling an age range that crossed 2 different GMFCS scales (<2 years and 2–4 years). Children’s GMFCS level may be reclassified after their second birthday, which may affect comparisons across the sample. The most significant limitation in all studies of feeding in young children remains the lack of a gold standard or consensus in the definition for the construct of OPD. A large range was found in the identification of OPD cases using each of the OPD measures, particularly between the SOMA and DDS. The SOMA was designed to detect clinically significant OPD, and therefore may lack sensitivity in detecting mild OPD. Conversely, although the DDS and pharyngeal signs appear to be detecting the milder feeding difficulties, these measures may be misclassifying behaviors as OPD that are present in young typically developing children. Although normative data exist for feeding efficiency28,29 and parent-reported acquisition of a limited number of oral behaviors,30 our future work assessing a typically developing reference sample with the SOMA and DDS will address some of the questions surrounding the validity of measures. Triangulation of videofluoroscopy swallow study results with clinical pharyngeal signs will further validate these findings, and will be the subject of future articles.

**CONCLUSIONS**

This study proposes a more plausible OPD estimate of 85% to reflect the prevalence of OPD in young children with CP based on direct clinical measures with reported validity and reliability, and a representative population-based sample across the range of gross motor functioning.

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**TABLE 5** Relationship Between Motor Capacity on the GMFM and Prevalence of OPD

<table>
<thead>
<tr>
<th>OPD</th>
<th>Odds Ratio</th>
<th>95% Confidence Interval</th>
<th>P Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>GMFM-66 overallb</td>
<td>0.89</td>
<td>0.84–0.95</td>
<td>.001</td>
</tr>
<tr>
<td>18–24 mo</td>
<td>0.90</td>
<td>0.81–1.00</td>
<td>.054</td>
</tr>
<tr>
<td>30–36 moa</td>
<td>0.93</td>
<td>0.88–0.99</td>
<td>.017</td>
</tr>
<tr>
<td>GMFM-88 (A) lying, rollingb</td>
<td>0.96</td>
<td>0.92–0.99</td>
<td>.020</td>
</tr>
<tr>
<td>18–24 mo</td>
<td>0.95</td>
<td>0.89–1.01</td>
<td>.126</td>
</tr>
<tr>
<td>30–36 moa</td>
<td>0.97</td>
<td>0.93–1.01</td>
<td>.121</td>
</tr>
<tr>
<td>GMFM-88 (B) sittingb</td>
<td>0.92</td>
<td>0.87–0.97</td>
<td>.004</td>
</tr>
<tr>
<td>18–24 mo</td>
<td>0.95</td>
<td>0.88–1.01</td>
<td>.078</td>
</tr>
<tr>
<td>30–36 moa</td>
<td>0.90</td>
<td>0.82–0.98</td>
<td>.017</td>
</tr>
<tr>
<td>GMFM-88 (C) crawling, kneelingb</td>
<td>0.96</td>
<td>0.94–0.99</td>
<td>.002</td>
</tr>
<tr>
<td>18–24 mo</td>
<td>0.98</td>
<td>0.95–1.01</td>
<td>.118</td>
</tr>
<tr>
<td>30–36 moa</td>
<td>0.95</td>
<td>0.91–0.99</td>
<td>.015</td>
</tr>
<tr>
<td>GMFM-88 (D) standingb</td>
<td>0.96</td>
<td>0.94–0.98</td>
<td>.001</td>
</tr>
<tr>
<td>18–24 mo</td>
<td>0.97</td>
<td>0.94–1.00</td>
<td>.050</td>
</tr>
<tr>
<td>30–36 moa</td>
<td>0.95</td>
<td>0.92–0.99</td>
<td>.013</td>
</tr>
<tr>
<td>GMFM-88 (E) walking, running, jumpingb</td>
<td>0.96</td>
<td>0.94–0.98</td>
<td>.000</td>
</tr>
<tr>
<td>18–24 mo</td>
<td>0.95</td>
<td>0.92–1.00</td>
<td>.027</td>
</tr>
<tr>
<td>30–36 moa</td>
<td>0.96</td>
<td>0.93–0.99</td>
<td>.007</td>
</tr>
</tbody>
</table>

a Crude odds ratios reported as covariates of postural instability and position predict perfectly for outcome.
b Statistically significant.

---

**FIGURE 2**

Relationship between DDS raw score and GMFM-66. Key: Pearson’s correlation: $r = -0.85$, $r^2 = 0.73$, $P < .000$, 18–24-month subgroup $r = -0.85$, $r^2=0.72$, $P < .000$, 30–36-month subgroup $r = -0.83$, $r^2 = 0.68$, $P < .000$. 

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motor severity levels. This study has confirmed previous findings, that OPD is related to gross motor severity, using a universally recognized gross motor classification (GMFCS). OPD was present across all GMFCS levels, which highlights the need for proactive screening of all young children with CP, even those from GMFCS I, to detect children at risk for feeding-related growth, nutrition, and respiratory compromise. To better understand the nature of OPD in this group of children, the OPD measures need further testing of their psychometric properties, particularly with reference to a typically developing sample. In addition, studies highlighting the specific impairments of children during the oral and pharyngeal phases of the swallow, and longitudinally during maturation of oral sensorimotor skills, will enable clinicians and researchers to better design and target interventions for children with feeding difficulties.

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REFERENCES


**TABLE 6 Relationship Between OPD and Motor Type and Distribution**

<table>
<thead>
<tr>
<th>OPD Overall, n (%)</th>
<th>Odds Ratio (95% Confidence Interval)⁎</th>
<th>P Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Spastic unilateral (base) 27 (75.0)</td>
<td>---</td>
<td>---</td>
</tr>
<tr>
<td>Spastic diplegia 22 (71.0)</td>
<td>0.8 (0.3-2.4)</td>
<td>.711</td>
</tr>
<tr>
<td>Spastic quadriplegiab 37 (100.0)</td>
<td>25.9 (2.4-154.8)</td>
<td>.002</td>
</tr>
<tr>
<td>Dyskinesia 6 (100.0)</td>
<td>4.5 (0.2-98.4)</td>
<td>.963</td>
</tr>
<tr>
<td>Hypotonia/ ataxia 10 (100.0)</td>
<td>7.3 (0.4-154.8)</td>
<td>.525</td>
</tr>
</tbody>
</table>

⁎ Crude odds ratios reported as covariates of postural instability and position predict perfectly for outcome.

b Classification includes triplegia (ie, bilateral spasticity with >2 limbs involved).

c Exposure predicts outcome perfectly, therefore calculated in episheet, based on Fisher’s Exact Test.


APPENDIX  Signs Suggestive of Pharyngeal Phase Impairment

- Gags when eating or drinking.
- Coughs when eating or drinking.
- Chokes when eating or drinking.
- Vomits when eating or drinking.
- Clears his/her throat often during or after meals.
- Needs to swallow a number of times to clear each mouthful of food or drink.
- Wheezes during/after eating or drinking.
- Has "stridor" when breathing in or out during eating or drinking.
- Becomes breathless and breathes quickly during eating or drinking.
- Breathing becomes labored or effortful during eating or drinking.
- Has a “rattly chest” after eating or drinking.
- Gets a “snuffy nose” after eating or drinking.
- Has a “gurgly voice” after eating or drinking.
- Has wet or “gurgly” breathing during or after eating or drinking.
- Has runny eyes or “eye tearing” after swallows of certain food or drinks.
- Seems to go “blue” around the lips/face or turn “dusky” or pale after drinking or eating.
- Generally refuses to eat or drink some food or fluid textures.
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