Oropharyngeal Dysphagia and Cerebral Palsy

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abstract

OBJECTIVES: To determine the progression of oropharyngeal dysphagia (OPD) in preschool-aged children with cerebral palsy (CP) according to gross motor function. It was hypothesized that fewer children would have OPD at 60 months compared with 18 to 24 months (predominately Gross Motor Function Classification System [GMFCS] I–II).

METHODS: Longitudinal population-based cohort of 179 children (confirmed CP diagnosis, born in Queensland in 2006–2009, aged 18–60 months at study entry [mean = 34.1 months ± 11.9; 111 boys; GMFCS I = 46.6%, II = 12.9%, III = 15.7%, IV = 10.1%, and V = 14.6%]). Children had a maximum of 3 assessments (median = 3, total *n* = 423 assessments). OPD was classified by using the Dysphagia Disorders Survey part 2 and rated from video by a certified pediatric speech pathologist. GMFCS was used to classify children's gross motor function.

RESULTS: OPD prevalence reduced from 79.7% at 18 to 24 months to 43.5% at 60 months. There were decreasing odds of OPD with increasing age (odds ratio [OR] = 0.92 [95% confidence interval (CI) 0.90 to 0.95]; $P < .001$) and increasing odds with poorer gross motor function (OR = 6.2 [95% CI 3.6 to 10.6]; $P < .001$). This reduction was significant for children with ambulatory CP (GMFCS I–II, OR = 0.93 [95% CI 0.90 to 0.96]; $P < .001$) but not significant for children from GMFCS III to V (OR [III] = 1.0 [95% CI 0.9 to 1.1]; $P = .897$; OR [IV–V] = 1.0 [95% CI 1.0 to 1.1]; $P = .366$).

CONCLUSIONS: Half of the OPD present in children with CP between 18 and 24 months resolved by 60 months, with improvement most common in GMFCS I to II. To more accurately detect and target intervention at children with persisting OPD at 60 months, we suggest using a more conservative cut point of 6 out of 22 on the Dysphagia Disorders Survey for assessments between 18 and 48 months.

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WHAT'S KNOWN ON THIS SUBJECT: The preschool years are a period of feeding skill acquisition and consolidation. There are only 2 longitudinal studies of dysphagia in children with cerebral palsy (from more limited age ranges), which found minimal changes to dysphagia prevalence and severity.

WHAT THIS STUDY ADDS: Prevalence of dysphagia halved between 18 and 60 months, and reduced most in children with ambulatory cerebral palsy. A Dysphagia Disorders Survey cut point of 6 out of 22 should be used in young children to better detect persisting dysphagia.

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Cerebral palsy (CP) is characterized by impaired development of motor skills, and much has been written regarding the progression and prognosis of gross motor function in this population.[1–4] Present in 2 out of 3 children with CP, oropharyngeal dysphagia (OPD) is known to influence their nutritional status, respiratory health, and parental stress.[5–8] Despite this, OPD has had limited attention, and little is known about its progression.[9,10] To inform management of OPD and its health consequences, and develop appropriate interventions influencing the nutritional and respiratory health trajectories for children with CP, it is important to document the early natural history of OPD.

The preschool years are a period of rapid oral sensorimotor development and consolidation, and they lay an important foundation for later skill acquisition.[11] By 18 months, children can typically manage a full range of food and fluid textures and drink from a standard cup.[11] Apart from our preliminary results,[10] there is only one other longitudinal study of feeding skill acquisition in children with CP. Researchers followed a cohort of 23 children from 4 to 7 years, and found a reduction in coughing across time, but no other changes to prevalence of impaired ingestion functions.[12] The study sample was small, and data were gathered from parental report on an informal feeding questionnaire. Consequently, there is limited knowledge of the progression of OPD from 18 to 60 months. Our team has published comprehensive preliminary data on the progression of OPD between 18 to 24 and 36 months in a population-based cohort of children with CP; however, final analysis is now presented for this cohort from 18 to 60 months (according to gross motor function). It is hypothesized that fewer children will have OPD at 60 months compared with 18 to 24 months, and these changes will predominately be observed in children with ambulatory CP.

METHODS

This is a longitudinal population-based study of a preschool-aged birth cohort with CP assessed from April 2009 to March 2015. Data were collected in 2 concurrent longitudinal studies, Queensland CP Child: Brain Structure and Motor Development (n = 227)[13] and Queensland CP Child: Growth, Nutrition and Physical Activity (n = 179).[14] Preliminary data from 53 children (aged 18–24 months at initial appointment and 36 months at follow-up) have been published previously.[10] All families gave written informed consent to participate. Ethics was approved through the University of Queensland Medical Research Ethics Committee (2008002260) and the Children's Health Services District Ethics Committee (HREC/08/QRCH/112).

Patients

The inclusion criteria were a confirmed diagnosis of CP and birth in Queensland between September 2006 and December 2009. Children with neurodegenerative conditions were excluded. All children were aged 18 to 60 months when assessed and could enter the study after diagnosis at any time in the study age range, with a semistandardized schedule of up to 3 assessments.

OPD Outcomes

The Dysphagia Disorders Survey (DDS): Pediatric (part 2) was selected as the primary OPD outcome after comprehensive systematic review and further testing of the psychometric properties.[15,16] The DDS evaluates 8 ingestion functions related to oral, pharyngeal, and esophageal phase dysphagia for children aged >18 months with CP or developmental disabilities.[9,17] The raw score (maximum = 22) indicates an individual's functional eating competency and has been used previously as a measure of OPD.[18] Primary validation and reliability was conducted in adults with developmental disability (mean = 33 years) and was shown to be strong.[17,19] The Pediatric version of the DDS was the tool-author's modification to the original tool (containing all the same items with the exception of item 1, part 1)[17] and has been used and validated in children from 18 months, and reproducibility was also shown to be strong.[9,18] Our team developed modified cut points for children with CP aged 18 to 36 months (as the standard scoring was found to overclassify OPD in this age range when applied to a sample of children with typical development). These modified cut points were used in this study for OPD classification of children aged 18 to 36 months.[9] To avoid overclassification based on measurement error in children aged 42 to 60 months, a cut point based on smallest detectable change (SDC) was calculated in the present analysis and subsequently applied for OPD classification in this age range.

An additional 16 clinical signs suggestive of pharyngeal phase impairment were included in the direct assessment because of limited evaluation of these signs on the DDS. Pharyngeal phase impairment was inferred if the child demonstrated any 1 of 16 signs (excluding a single cough on thin fluids in children <36 months)[20] and was rated live and from video by the speech pathologist.

Children's eating and drinking abilities were classified according to 5 levels on the Eating and Drinking Ability Classification System (EDACS).[21] The EDACS was administered to children >3 years as a result of its validation age. The EDACS has strong intrarater (intraclass correlation coefficient [ICC] = 0.95) and fair to strong interrater reliability between professionals (ICC = 0.79–0.93).[21,22]

The CP Child Feeding Questionnaire is a parent-completed questionnaire designed specifically for this study.[10] The items analyzed in the current study were parent-reported severity of eating and drinking (10-point visual analog scales), textures included in the child's diet, feeding method (degree of oral supplementation and feeding tubes, 1–5), challenging behaviors (total of 16), and stress during mealtimes or worry of child's growth (two 5-point Likert scales).

Motor Outcomes and Covariates

Children's gross motor function was classified according to 5 levels on the Gross Motor Function Classification System (GMFCS). At 4 to 6 years, GMFCS I to II describe independently ambulating children, GMFCS III describes those requiring walking assistance, and GMFCS IV to V describe those requiring wheeled mobility.[2] The <2, 2 to 4, and 4- to 6-year-old versions were used in the current study. Children's manual ability was classified according to 5 levels on the Manual Ability Classification System (MACS),[23] with reproducibility in our cohort previously shown to be moderate.[24] Motor type and distribution were classified on the Surveillance of CP in Europe.[25] The covariates of epilepsy and preterm birth were reported by parents during the initial physician interview by using the physician checklist designed for this study.[13]

Procedures

Children attended the hospital for mealtime and gross motor assessment. A digital video recording of a standardized mealtime evaluation was collected by a trained research assistant, with children well positioned in their typical mealtime seating and using their regular utensils. Three presentations of 4 textures (puree, lumpy, chewable, and fluid) were presented by the primary carer,[26] followed by the child completing their snack as usual. Mealtime evaluations were rated by a pediatric speech pathologist with 13 years of experience in pediatric disability (DDS certified). All motor ratings were conducted by 2 trained physiotherapists.

Reproducibility Study

OPD reproducibility in this article complements existing reproducibility completed by our team for children aged 18 to 36 months.[9,24] Forty children (4 per GMFCS level, stratified for age bands 48 and 60 months) were selected randomly by an independent researcher for analysis of intrarater and interrater reproducibility of the DDS and clinical signs. Independent speech pathologists (certified in the DDS) rated the videos and were blinded to reproducibility case status. Intrarater reproducibility ratings were performed >2 weeks after initial ratings. Reproducibility of the GMFCS between the 2 experienced physiotherapists in this cohort was analyzed for an independently selected random sample of 284 ratings.

Statistical Analysis

Demographic data were presented with descriptive statistics (initial assessment and 60 months), and the sample was compared with the Australian CP Register[27] to determine representativeness by using a χ^2 test for trend. Intra- and interrater reproducibility were assessed by using Cohen's Kappas, percentage agreement ("perfect" and "close" indicating 1 level on either side), and ICCs (2-way mixed effects, individual). The SE of measurement and SDC were calculated from the intrarater reliability. Univariate regression analyses were undertaken for OPD risk factors (age, sex, preterm status, epilepsy, motor type). Variables consistently significant at the $P = .05$ level were then included in all multivariate regressions. The relationships between OPD outcomes at 60 months and (1) GMFCS and (2) MACS were determined with logistic, ordered, and linear regression. The longitudinal analysis of OPD classification and severity was conducted by using multilevel mixed effects logistic (OPD classification) and linear regression (OPD severity), with interaction terms between age and GMFCS. The child's identification number was entered as a random effect for all mixed effect models. The trajectory of OPD was classified for children with 3 assessment points, and its association with GMFCS was determined by using ordered logistic regression. The prediction of earlier OPD outcomes for "persistent OPD" (at 60 months) was analyzed by using sensitivity and specificity and receiver operating characteristic (ROC) curves. All data analyses were performed by using Stata 13.1 (StataCorp, College Station, TX).

RESULTS

Sample Characteristics

There were 244 eligible children referred to the study, with 179 children participating and a total of 423 data points (Supplemental Fig 3). Sample characteristics are shown in Table 1 and are considered representative of the Australian CP population on the basis of sex ($P = .243$ for initial assessment, $P = .372$ for 60 months), GMFCS ($P = .001$ for initial assessment, $P = .216$ for 60 months), and motor type ($P = .499$ for initial, $P = .800$ for 60 months).

Reproducibility Study

The intrarater reproducibility of the DDS in children 48 to 60 months was found to be excellent ($\kappa = 0.94$, $P < .001$), and the intrarater reproducibility of clinical signs suggestive of pharyngeal phase impairment was substantial to excellent ($\kappa = 0.7–1.0$, $P < .001$) (Supplemental Table 3). The interrater reproducibility was similarly strong for the DDS ($\kappa = 0.82$, $P < .001$) and clinical signs ($\kappa = 0.39–0.84$, $P < .001$). The SE of measurement for DDS scores for children aged 48 to 60 months was 0.6 and for SDC was 1.7. A cut point of

TABLE 1 Characteristics of Preschool-Aged Children With CP in the Longitudinal OPD Study, Initial Assessment (n = 179) and 60 Months Assessment (n = 135)

	Initial Assessment	60 mo Assessment
Age in months, mean (SD)	34.1 (11.9)	61.4 (2.7)
Sex (male), *n* (%)	111 (62.0)	82 (60.7)
GMFCS level, *n* (%)		
I	84 (46.9)	60 (44.4)
II	23 (12.9)	29 (21.5)
III	28 (15.6)	18 (13.3)
IV	18 (10.1)	11 (8.2)
V	26 (14.5)	17 (12.6)
MACS level, *n* (%)		
I	73 (40.8)	76 (56.3)
II	59 (33.0)	29 (21.5)
III	14 (7.8)	10 (7.4)
IV	12 (6.7)	3 (2.2)
V	21 (11.7)	17 (12.6)
Primary motor type, *n* (%)		
Spasticity	157 (87.7)	117 (86.7)
Dyskinesia	8 (4.5)	8 (5.9)
Ataxia	9 (5.0)	7 (5.2)
Hypotonia	5 (2.8)	3 (2.2)
Motor distribution, *n* (%)		
Monoplegia or hemiplegia	62 (34.6)	50 (37.0)
Diplegia	44 (25.0)	37 (27.4)
Triplegia or quadriplegia	73 (40.8)	48 (35.6)
Preterm birth (<37 wk), *n* (%)	82 (45.8)	60 (44.4)
Epilepsy, *n* (%)	30 (24.4)	24 (27.3)
Tube feeding, *n* (%)	19 (10.6)	16 (11.9)

n = 11 children's initial assessments (2.5%) were at 60 mo; these children's data have been presented in both columns. GMFCS reclassification was 28.3% (n = 145 repeated classifications).[28] MACS reclassification was 35.4%. Epilepsy data were missing for n = 56 children on initial assessment and for n = 47 children at 60 mo.

≥2 was used for all OPD classifications of children aged 42 to 60 months described in the subsequent analysis. Interrater reliability of the GMFCS was also found to be strong (κ = 0.83, P < .001) (Supplemental Table 4).

OPD Characteristics According to Motor Classification Systems: 60-Month Cross-Sectional Data

Almost all OPD outcomes were related to GMFCS, including prevalence and severity of OPD, EDACS, textures included in children's diets, and feeding method (Table 2). OPD outcomes that were not related to GMFCS (after adjusting for other sample characteristics) were the number of challenging behaviors and self-reported parental stress or worry during mealtimes, which were significantly associated with each other. A secondary analysis found a strong significant association between primary OPD outcomes and MACS (Supplemental Table 5).

Progression of OPD Classification and Severity

The proportion of children with OPD (on the DDS) decreased stepwise from 79.7% at 18 to 24 months to 43.5% at 60 months, as shown in Fig 1. The odds of children having OPD decreased with increasing age (odds ratio [OR] = 0.92 [95% confidence interval (CI) 0.90 to 0.95]; P < .001) and increased with increasing GMFCS level (OR = 6.2 [95% CI 3.6 to 10.6]; P < .001). The interaction between collapsed GMFCS and age indicated no significant reduction in OPD for children from GMFCS III (OR = 1.0 [95% CI 0.9 to 1.1]; P = .897) or IV to V (OR = 1.0 [95% CI 1.0 to 1.1]; P = .366) relative to GMFCS I to II. The reduction for ambulatory children with CP was significant (GMFCS I–II, OR = 0.93 [95% CI 0.90 to 0.96]; P < .001).

On average, children's DDS scores decreased by 0.05 points for each month increase in age (β = −.05, P < .001) (Fig 2). OPD trajectories were significantly related to GMFCS (Table 2), although this was no longer significant when adjusted for sex, preterm status, and epilepsy. Children from GMFCS I generally had no OPD or a trajectory of improvement, with 3 children declining and 2 fluctuating. Children from GMFCS IV either maintained a classification of OPD or maintained a classification of no OPD across the 3 assessment points, and all children from GMFCS V remained classified as OPD from initial assessment.

Prediction of OPD at 60 Months

All early assessments were excellent predictors of persisting OPD (18–24 months ROC = 0.92 [95% CI 0.84 to 0.99]; 30–36 months ROC = 0.96 [95% CI 0.91 to 1.0]; 42–48 months ROC = 0.80 [95% CI 0.68 to 0.92]), with the best prediction from the 30 to 36 month assessment. A DDS score ≥6 was the most accurate cut point for persistent OPD at 60 months, accurately classifying 84.8% of persistent OPD from 18 to 24 months (sensitivity = 84.2, specificity = 85.1), 90.2% from 30 to 36 months (sensitivity = 81.1, specificity = 96.4), and 91.7% from 42 to 48 months (sensitivity = 82.6, specificity = 97.3).

DISCUSSION

To our knowledge, with this study, we are the first to document the prevalence and progression of OPD in a large, prospective, population-based cohort representative of preschool-aged children with CP. OPD prevalence halved between 18 to 24 and 60 months, and this was particularly evident in children with ambulatory CP. Direct clinical assessment of OPD was conducted by using valid and reliable tools, which gives confidence in the findings.

OPD in Children With CP Aged 60 Months

Consistent with our previous work,[20,24,29,30] children with poorer

TABLE 2 Feeding Characteristics of Children Aged 60 Months With CP in the Longitudinal OPD Study by GMFCS (n = 135)

	Overall, n (%)	GMFCS I, n (%): n = 60	GMFCS II, n (%): n = 29	GMFCS III, n (%): n = 18	GMFCS IV, n (%): n = 11	GMFCS V, n (%): n = 17	Crude OR (95% CI); P	Adjusted OR (95% CI); P
OPD classification overall	78 (57.8)	19 (31.7)	18 (62.1)[a]	15 (83.3)[a]	9 (81.8)[a]	17 (100.0)[a]	3.0 (1.9 to 4.6); <.001	3.3 (1.7 to 6.3); <.001
DDS (modified)	58 (43.0)	8 (13.3)	12 (41.4)[a]	12 (66.7)[a]	9 (81.8)[a]	17 (100.0)[a]	3.7 (2.4 to 5.7); <.001	4.4 (2.2 to 8.8); <.001
Pharyngeal phase signs	41 (30.4)	2 (3.3)	7 (24.1)[a]	6 (33.3)[a]	9 (81.8)[a]	17 (100.0)[a]	4.8 (2.9 to 7.9); <.001	9.0 (3.3 to 25.0); <.001
Parent report	58 (45.0)	13 (22.8)	9 (34.6)	12 (66.6)[a]	7 (63.6)[a]	17 (100.0)[a]	2.5 (1.8 to 3.6); <.001	2.2 (1.4 to 3.5); .001
OPD severity, mean (95% CI)	4.3 (3.1 to 5.5)	0.5 (0.1 to 0.9)	1.8 (0.8 to 2.7)	4.4 (2.2 to 6.6)[a]	8.3 (4.4 to 12.2)[a]	20.2 (19.0 to 21.4)[a]	β = 4.0 (3.7 to 4.3); <.001	β = 3.9 (3.3 to 4.5); <.001
OPD trajectory (on DDS)							1.4 (1.1 to 1.8); .010	1.3 (0.9 to 1.8); .233
Unchanged (no OPD)	41 (43.2)	30 (69.8)	5 (41.7)	4 (21.1)	1 (16.7)	0 (0.0)	—	—
Unchanged (OPD)	33 (34.7)	3 (7.0)	2 (16.7)	9 (47.4)	5 (83.3)	15 (100.0)	—	—
Improved	12 (12.6)	8 (18.6)	2 (16.7)	2 (10.5)	0 (0.0)	0 (0.0)	—	—
Declined	3 (3.2)	0 (0.0)	1 (8.3)	2 (10.5)	0 (0.0)	0 (0.0)	—	—
Fluctuated	6 (6.3)	2 (4.7)	2 (16.7)	2 (10.5)	0 (0.0)	0 (0.0)	—	—
EDACS							6.1 (4.0 to 9.2); <.001	8.2 (4.2 to 15.7); <.001
I	79 (58.5)	55 (91.7)	16 (55.2)	6 (33.3)	2 (18.2)	0 (0.0)	—	—
II	25 (18.5)	3 (5.0)	12 (41.4)	7 (38.9)	3 (27.3)	0 (0.0)	—	—
III	12 (8.9)	2 (3.3)	0 (0.0)	5 (27.8)	3 (27.3)	2 (11.8)	—	—
IV	7 (5.2)	0 (0.0)	1 (3.4)	0 (0.0)	3 (27.3)	3 (17.7)	—	—
V	12 (8.9)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	12 (70.5)	—	—
Support for feeding							8.8 (4.8 to 16.2); <.001	9.6 (4.0 to 22.9); <.001
Independent	97 (71.9)	57 (95.0)	28 (96.6)	10 (55.6)	2 (18.2)	0 (0.0)	—	—
Partial	19 (14.1)	3 (5.0)	1 (3.4)	7 (38.9)	8 (72.3)	0 (0.0)	—	—
Total	19 (14.1)	0 (0.0)	0 (0.0)	1 (5.6)	1 (9.1)	17 (100.0)	—	—
Textures included								
Purees	114 (89.1)	53 (94.6)	25 (96.2)	17 (94.4)	11 (100.0)[a]	8 (47.1)[a]	0.5 (0.3 to 0.7); <.001	0.5 (0.4 to 0.7); <.001
Semisolids	112 (87.5)	55 (98.2)	25 (96.2)	17 (94.4)	9 (81.8)[a]	6 (35.3)[a]	0.3 (0.2 to 0.5); <.001	0.4 (0.3 to 0.5); <.001
Chewables	113 (88.3)	56 (100.0)	26 (100.0)	18 (100.0)	10 (90.9)	3 (17.7)[a]	0.0 (0.0 to 0.2); <.001	0.1 (0.0 to 0.2); <.001
Tough chewables	100 (78.1)	52 (92.9)	23 (88.5)	16 (88.9)	8 (72.7)	1 (5.9)[a]	0.4 (0.4 to 0.5); <.001	0.4 (0.3 to 0.6); <.001
Thin fluids	114 (89.1)	56 (100.0)	26 (100.0)	17 (94.4)	11 (100.0)	4 (23.5)[a]	0.1 (0.0 to 0.3); <.001	0.1 (0.0 to 0.2); <.001
Thick fluids	4 (3.1)	0 (0.0)	0 (0.0)	1 (5.6)	0 (0.0)	3 (42.9)	5.7 (1.5 to 21.8); .011	18.4 (0.9 to 393.2); .062
Feeding method (n = 128)							3.2 (2.6 to 3.9); <.001	3.8 (0.6 to 22.8); <.001
Full oral	112 (87.5)	57 (100.0)	25 (100.0)	15 (83.3)	11 (100.0)	4 (23.5)	—	—
Partial tube	9 (7.0)	0 (0.0)	0 (0.0)	3 (16.7)	0 (0.0)	9 (52.9)	—	—
Full tube	7 (5.5)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	7 (41.2)	—	—
Challenging behaviors (16), mean (95% CI)	4.1 (3.3 to 4.9)	3.8 (2.7 to 4.9)	3.7 (2.1 to 5.4)	3.6 (1.4 to 5.8)	6.4 (3.1 to 9.6)[a]	4.7 (1.7 to 7.7)	β = .2 (−0.1 to 0.6); .141	β = .3 (−0.2 to 0.7); .219
Parent stress, mean (95% CI)	1.6 (1.3 to 1.8)	1.6 (1.4 to 1.8)	1.5 (0.6 to 2.4)	1.6 (1.2 to 2.0)	2.2 (1.5 to 2.9)[a]	0.9 (0.0 to 2.3)[a]	1.1 (1.0 to 1.3); .088	0.9 (0.6 to 1.3); .458
Parent worry about growth, mean (95% CI)	1.7 (1.4 to 1.9)	1.6 (1.4 to 1.9)	1.5 (0.6 to 2.4)	1.7 (1.2 to 2.1)	2.0 (1.5 to 2.4)[a]	1.8 (1.3 to 2.2)[a]	1.2 (1.1 to 1.4); .224	1.1 (0.8 to 1.5); .681

Parent stress was significantly associated with number of challenging behaviors (adjusted β = 3.0 [95% CI 2.6 to 3.3], P < .001); adjusted for age, sex, preterm status, and epilepsy. Trajectory data include only children with full longitudinal data (3 data points), n = 95. —, not applicable or not available.

[a] Indicates individual GMFCS levels that are significantly different compared with GMFCS I.

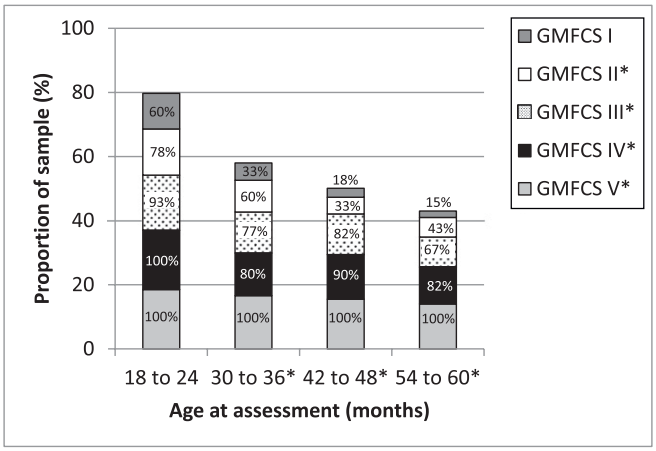

FIGURE 1

Proportion of children with OPD (on the DDS) according to gross motor function and assessment type. The figure shows the relative contribution of GMFCS level to overall proportion with OPD at each assessment age. The percentages within the bar segments reflect the percentage of GMFCS level with OPD. Data points displayed do not account for multiple records. * Indicates age (OR = 0.92, [95% CI 0.90 to 0.95]; $P < .001$) and GMFCS (OR = 6.2 [95% CI 3.6 to 10.6]; $P < .001$) were significantly related to OPD outcome on multilevel mixed effects logistic regression.

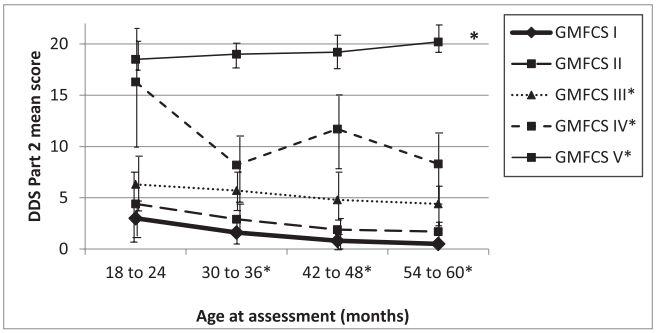

FIGURE 2

The average decrease per month was 0.05 DDS points (β = −.05, $P < .001$); GMFCS I β = −.06, $P < .001$. Relative to children classified as GMFCS I, scores were significantly higher for those from GMFCS III to V. For the interaction between GMFCS and age for GMFCS V (compared with GMFCS I), $P = .002$. Data points displayed do not account for multiple records. * Indicates variables significant at $P < .05$, including interaction between GMFCS and age (significant for GMFCS V).

gross motor function on the GMFCS and upper limb function on the MACS[24] had an increased presence of almost all OPD outcomes. This association between OPD outcome and poorer motor function has been widely reported,[5,18,31–34] reflecting the severity of the brain lesion. The number of challenging child behaviors and self-reported parent stress during mealtimes were strongly related to one another, but these were most commonly reported in children from GMFCS III to IV. This finding is in contrast to the Oxford Feeding Study, in which researchers found 20% of parents reported feeding as stressful or unenjoyable, with increased rates of stress with increasing gross motor severity ($P < .005$).[5] The Oxford study only recruited children with reported swallowing or articulation difficulties, which may have affected the finding.

Progression of OPD Classification and Severity in the Preschool Years for Children With CP

As children with CP got older, rates of OPD decreased. The literature reports that by 18 months, children can manage a full range of food and fluid textures, although skill development for more complex textures continues beyond 6 years.[11,35] On the basis of the literature describing typical progression of feeding abilities, the rapid decrease in prevalence between 18 to 24 and 30 to 36 months found in this study was expected. It is important to consider, however, that the modified cut points for children aged 18 to 36 months were used to classify OPD,[9] which should account for "developmental limitations to ingestion functions." In contrast to the typical progression of feeding, we documented a continued decrease in OPD prevalence throughout the preschool period in children with CP, which may correspond to delayed rather than disordered feeding abilities in some of these children. Our findings were also in contrast to the other longitudinal study of children with CP aged 4 to 7 years, in which researchers noted a decrease only in coughing in children whose feeding abilities were considered within normal limits.[12] Their study size of 23 children was small and heterogenous, which reduces interpretation of these findings.

Consistent with children's clinical profile at 60 months, the strong relationship between increasing OPD prevalence and poorer gross motor function (II–V) persisted as children aged. In particular, children classified as GMFCS I had the greatest decrease in prevalence (from 60% to 15%), and this was also noted in the trajectory analysis (these children generally had no OPD to begin with or they improved). Interestingly, the prevalence of OPD increased for children from GMFCS III to IV between 30 to 36 and 42 to 48 months. The pattern of a period of decline in the midrange GMFCS levels was also noted in our earlier longitudinal work, which we attributed to a later introduction of more complex food textures and fluid utensils.[10] This could also be a factor in the current study, although it could also be related to the precision of the estimates. The trajectory for children from GMFCS IV

was consistently unchanging, either maintaining OPD classification from first to final assessment or maintaining no OPD. All children from GMFCS V had OPD classified at each assessment. These findings suggest that the developmental feeding trajectories for children from GMFCS IV to V are set by 18 to 24 months, whereas skills may continue to change for children from GMFCS I to III.

The severity of OPD followed a similar rate of improvement for children from GMFCS I, II, and III, with an average change of 2 to 3 DDS points between 18 to 24 and 60 months (equating to improvement of 2–3 ingestion functions). From our previous work,[9] this could reflect an improvement in developmental skills like biting firm foods and sipping without spilling from an open cup. Children from GMFCS IV had the largest variation in scores, resulting in a less predictable progression of OPD severity. In previous cross-sectional analysis, this subgroup had the largest range in DDS scores,[29] but this may also be related to fewer children classified as GMFCS IV. Children from GMFCS V were the only subgroup who had a decline in DDS scores by 60 months, which in part relates to the introduction or increase in tube feeding. Children who were completely tube fed did not have a feeding evaluation completed, so we cannot conclude whether the increase in tube feeding was related to a decline in oropharyngeal abilities, to parents' increased acceptance of tube feeding, or other factors.

Prediction of Persistent OPD in Children With CP

An understanding of the varied patterns and trajectories of OPD for children based on their GMFCS level is important to inform the decision-making process for clinicians. This study found early DDS assessments (from as young as 18 months) were excellent predictors of persisting OPD at 60 months. By 36 months, the prediction of "persistent" OPD was optimized. To more accurately detect and target intervention for children with persisting OPD at 60 months, we suggest using a more conservative cut point of 6 out of 22 on the DDS for assessments between 18 and 48 months.

Limitations

This study is the first of its kind in which the progression of OPD in a large population-based sample is documented. These findings are expected to assist clinicians in understanding the course of OPD in the preschool years for a child with CP and contribute to improved management and prognostication. A major limitation to our findings was the lack of detailed intervention data. Without these data, we were unable to determine if changes were related to the provision of feeding interventions or reflective of the early natural history of CP. Another limitation of the current study was the absence of videofluoroscopic swallow data to contribute diagnostic information regarding the pharyngeal phase of the swallow. Results of children who had a videofluoroscopic swallow indicated clinically were reviewed, but there were insufficient numbers for statistical analysis, which meant pharyngeal phase impairment could only be inferred from clinical signs.

CONCLUSIONS

We showed that half of the OPD observed in children with CP across the full spectrum of motor severity between 18 and 24 months is likely to resolve by 60 months. To more accurately detect and target appropriate interventions toward children with persisting OPD at 5 years, a more conservative cut point of 6 out of 22 is recommended. Even beyond their third year, children with CP continued to show improvements to their feeding skills, particularly ambulatory children. As such, children from GMFCS I presenting with OPD should be monitored closely (for feeding efficiency and safety), and if indicated, oral sensorimotor treatments should be provided. OPD classification of children with nonambulatory CP is likely to remain unchanging from as young as 18 to 24 months, suggesting that multimodal compensatory strategies with emphasis on nutrition and respiratory health should be prioritized from this age. The findings from this study are expected to assist in improved screening and management of OPD in children with CP across the full spectrum of gross motor severity.

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ABBREVIATIONS

CI: confidence interval
CP: cerebral palsy
DDS: Dysphagia Disorders Survey
EDACS: Eating and Drinking Ability Classification System
GMFCS: Gross Motor Function Classification System
ICC: intraclass correlation coefficient
MACS: Manual Ability Classification System
OPD: oropharyngeal dysphagia
OR: odds ratio
ROC: receiver operating characteristic
SDC: smallest detectable change

supervision; and all authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work to ensure its accuracy and integrity.

This trial has been registered with the Australia New Zealand Clinical Trials Registry (trial registration ACTRN12611000616976).

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