# Communication disorders in young children with cerebral palsy

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**ABBREVIATIONS**

| | |
|---|---|
| AAC | Augmentative and alternative communication |
| CFCS | Communication Function Classification System |
| FCCS | Functional Communication Classification System |
| MACS | Manual Ability Classification System |
| SLT | Speech and language therapy |
| VSS | Viking Speech Scale |

**AIM** To test the prediction of communication disorder severity at 5 years of age from characteristics at 2 years for children with cerebral palsy (CP) whose communication is giving cause for concern.

**METHOD** In this cohort study, 77 children (52 males; 25 females) with communication difficulties and CP were visited at home at 2 (mean 2y 4mo; SD 3mo) and 5 (mean 5y 5mo; SD 4mo) years of age. Information on the type and distribution of motor disorder, seizures, gross and fine motor function, hearing, and vision were collected from medical notes. Non-verbal cognition, language comprehension, language expression, spoken vocabulary, and methods of communication were assessed directly at age 2 years. At 5 years, communication and speech function were rated using the Communication Function Classification System (CFCS), Functional Communication Classification System (FCCS), and Viking Speech Scale (VSS).

**RESULTS** In multivariable regression models, CP type, Gross Motor Function Classification System level, vision, the amount of speech understood by strangers, non-verbal cognition, and number of consonants produced at age 2 years predicted the CFCS level at age 5 years ($R^2$=0.54). CP type, Manual Ability Classification System level, amount of speech understood, vision, and number of consonants predicted the FCCS level ($R^2$=0.49). CP type, amount of speech understood by strangers, and number of consonants predicted the VSS level ($R^2$=0.50).

**INTERPRETATION** Characteristics at 2 years of age predict communication and speech performance at 5 years, and should inform referral to speech and language therapy.

Around 55% of children with cerebral palsy (CP) have difficulties engaging in face-to-face conversation.[1–4] Approximately 21% to 36% have dysarthria,[4,5] which limits the intelligibility of their speech, and 19% to 32% are unable to speak at all.[1,5,6] The risk of dysarthria and lack of speech is greater for children with dyskinetic than spastic forms of CP,[3,5,7] and for children born at term with a not unexpectedly high birthweight.[1] Magnetic resonance imaging studies show that lesions in cortical/subcortical regions and basal ganglia are associated with a greater risk of poor speech and communication outcomes than periventricular leukomalacia.[3,5,8] Speech and communication performance in childhood are both positively associated with oromotor function,[9] gross and fine motor function, and non-verbal cognition.[3,5,9–11] Language development appears highly correlated with non-verbal cognition.[11]

Recent studies have provided information that could guide prognosis. They show that communication performance (sending and receiving messages) is predicted by symbolic behaviour, speech, and gross motor function at 18 to 24 months,[2,10,12] and that receptive and expressive language outcomes are predicted by cognition and motor function from 3 years of age.[13] Children with CP who were established talkers at 2 years of age are likely to have (approaching) age-appropriate receptive and expressive language at 4 years.[14] Children who had no spoken words at 2 years of age are likely to be non-speaking,[15] and have slow receptive language development trajectories and language comprehension ages below their chronological age at 4 years.[14] Most children who are non-speaking at 2 years have severely impaired motor function (Gross Motor Function Classification System [GMFCS] level IV or V) and spastic-type motor disorder. Further research is needed to extend the profile of children's characteristics to inform prognostication.[14]

Previous research has identified early speech, language, cognition, and motor function as predictors of communication at around the time of school entry for population cohorts of children with CP. However, predictors may differ for the subgroup of children with CP who experience

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communication difficulties in early life. This paper examines which characteristics predict speech and communication at 5 years of age for children whose communication is already causing concern at 2 years, in order to inform prognostic messages to parents and invention pathways. We examined the predicative ability of all of the characteristics reported to be associated with communication outcomes in previous research, as no reported studies have used a full battery of robust assessments to examine both speech and language development, and cognitive and motor function. To inform referral decisions, we also examined the predictions using measures available to paediatricians and speech and language therapists separately.

## METHOD

### Study design

This was a prospective cohort study of 2-year-old children with communication difficulties and likely to have a CP diagnosis.

### Sample

We recruited 125 children born between 1st February 2005 and 31st December 2010, who had a confirmed diagnosis of CP or a nonprogressive motor disorder who were likely to receive a diagnosis of CP, and whose communication was giving a caregiver or health/social care provider cause for concern at 2 to 3 years of age. Children with hypotonia alone were excluded after Surveillance of Cerebral Palsy in Europe classification.[16] No other exclusions were applied. This paper reports the data from children who entered the study at 2 years of age.

Local clinicians identified children from their current caseloads, and referred families to the project. Clinicians also provided information on birth year, sex, type, and distribution (uni/bilateral) of motor disorders for children who were not referred to the study. The research was approved by Cambridgeshire 4 National Health Service Research Ethics Committee and all referring National Health Service Trusts (08H030571). The children's parents/guardians provided written consent.

### Predictor variables at age 2 years

Neurological/medical characteristics previously associated with motor, cognitive, or communication function were extracted from children's medical notes: gestational age at birth (in completed weeks);[1,12] birthweight in grams;[1] type and distribution of motor disorder[3] after the Surveillance of Cerebral Palsy in Europe classification (spastic, dyskinetic, and ataxic; unilateral or bilateral);[16] neonatal seizures (yes/no);[1,12] vision[1] (using the North of England Collaborative CP Survey: no impairment; impairment but not registered as blind/partially sighted on UK government register; or registered as severely visually impaired or blind); hearing acuity[1] (North of England Collaborative CP Survey: no problem/mild impairment; 41–70dB hearing loss; >70dB hearing loss); and additional diagnoses.

**What this paper adds**

- Data available to paediatricians on children with cerebral palsy at 2 years of age predicts their speech and communication at 5 years.
- This includes cerebral palsy type, a four-point rating of speech intelligibility, GMFCS level, MACS level, and vision.
- Adding detailed speech and language therapy measures slightly increases the precision of prediction.

A research speech and language therapist assessed children's functional performance during home visits. Motor function was rated using the GMFCS[17] and the Manual Ability Classification System (MACS).[18] The Mini-MACS was not available at the start of the study.[19] Non-verbal cognition was assessed using the visual reception scale of Mullen Scales of Early Learning.[20] All children included in the study had sufficient vision to attend to the objects in the assessment. In both the assessment of non-verbal cognition and receptive language (below), children with limited fine motor control (MACS levels IV and V) completed the assessments by eye/hand pointing toward objects and their intended destinations. Methods of response were examined in test trials before test administration. If the therapist was unsure of a child's response during test administration, the item was repeated. If a second response was unclear, the item was marked as failed. As the adaptations introduced further cognitive load to the assessment tasks, the results could represent conservative estimates of children's functioning. To characterize children's oral motor function, parents were asked about whether their child fed orally and if they had any difficulties eating and drinking (fed orally, no problem; fed orally but oral motor problem; fed orally plus nasogastric or [percutaneous] gastrostomy tube; or not fed orally); the Eating and Drinking Ability Classification System[21] was not available at the start of the study and is not validated for children at 2 years of age.

#### *Speech*

Speech was assessed as the number of different consonants observed in play with a parent during the visit and/or produced when naming pictures in the articulation subtest of the Diagnostic Evaluation of Articulation and Phonology test.[22] Single words and word approximations produced in play were often spontaneously repeated by parents, as confirmation of their understanding. The research therapist marked the consonants she perceived the child to produce in words or word approximations during play, and when naming pictures in the Diagnostic Evaluation of Articulation and Phonology test, on the case record form during the assessment visit. Assessments were audio recorded. Interrater reliability of consonant identification was assessed by a second, independent rater identifying consonants from 32 randomly selected audio recordings (one-way intraclass correlation coefficient 0.94, 95% confidence interval [CI] 0.89–0.97). Children who did not produce spoken words that were recognizable in context during interaction with their parent, and during assessments of non-verbal cognition and receptive language, were assessed as having no consonants; the Diagnostic Evaluation of Articulation and Phonology test was not attempted with these children.

Coplan and Gleason's four-point parental rating of how much of their child's speech understood by strangers (1, all or almost all; 2, three-quarters; 3, about one-half; and 4, less than one-half. Children who did not speak were rated at 4) was also used.[23]

### Language

The numbers of words in children's spoken vocabularies were assessed using the parent-completed MacArthur Communicative Development Inventory UK Edition,[24] and expressive and receptive language subscales of the Preschool Language Scales, Fourth Edition UK.[25]

### Classification of speech and communication and additional diagnoses at age 5 years

A research speech and language therapist visited the children at home when they were 5 years of age, to assess the development of their speech, language, and non-verbal cognition using standardized tests and to evaluate their expressive communication using a semi-scripted elicitation procedure developed for children who use augmentative and alternative communication (AAC).[26,27] Visits lasted approximately 90 minutes including short breaks. The therapist rated each child's communication and speech performance from observation of the child in these assessments, and in free play with their parents using their own toys. The Communication Function Classification System (CFCS)[28] categorizes each child's communication performance as both a sender and receiver of messages. The Functional Communication Classification Scale (FCCS)[29] rates a child's observable communication performance using speech, gesture, and/or AAC. The Viking Speech Scale (VSS)[30] grades children's speech intelligibility. Each scale is ordinal, with the highest levels (level I) indicating no difficulty with performance. Table 1 describes the classification systems.

At age 5 years, the research speech and language therapist also collected information about any additional diagnoses made since the case note review at 2 years.

**Table 1:** Summary descriptors for levels of the CFCS, FCCS, and VSS

| Level | CFCS | FCCS | VSS |
|---|---|---|---|
| I | Effective sender and receiver with familiar and unfamiliar partners | Effective communicator in most situations | No motor speech disorder |
| II | Effective, but slower-paced sender and/or receiver with familiar and/or unfamiliar partners | Effective communicator in most situations but may need help | Motor speech disorder but usually understandable to unfamiliar listeners |
| III | Effective sender and receiver with familiar partners | An effective communicator in some situations | Motor speech disorder and not usually understandable to unfamiliar listeners out of context |
| IV | Inconsistent sender and/or receiver with familiar partners | Assistance required in most situations, especially with unfamiliar partners | No understandable speech |
| V | Seldom effective sender and receiver, even with familiar partners | Communicates unintentionally using movements and behaviour | — |

CFCS, Communication Function Classification System; FCCS, Functional Communication Classification System; VSS, Viking Speech Scale.

### Analysis

As more severe impairments may be detected earlier, recruiting children at any point in their third year of life year risks introducing bias in recruitment, with children who have more severe impairments potentially being identified for the study earlier. We compared children seen at 24 to 29 months and 30 to 35 months of age on all measures, using Mann–Whitney $U$ tests for continuous measures and $\chi^2$ tests for categorical/ordinal variables, to assess if children differed in their severity of impairment. We measured the association between language domains and non-verbal cognition using Pearson's product moment correlations as data were not normally distributed. We also examined the concordance between the three communication classifications at 5 years using the weighted kappa test and agreement between raters on speech data using the intraclass correlation coefficient (one-way random effect).

Univariable ordinal logistic regression models, using the proportional odds assumption, examined the prediction of speech (VSS) and communication (CFCS and FCCS) at 5 years by demographic, early medical history, diagnostic category, and functioning at 2 years. Tables present the results of the regression models as proportional odds ratios, interpreted as an increase in the odds of being in a less functional CFCS, FCCS, or VSS level for each unit increase of the predictor variable, with corresponding 95% CIs. The significant ($p$<0.05) variables from the univariable models were entered into the multivariable ordinal logistic regression models in order of reducing significance. Likelihood ratio tests examined whether additional categorical and ordinal variables were significant within the multivariable models ($p$<0.05). Separate models tested data available to paediatricians (demographic, medical history, and performance measures including rating of speech at 2 years) and speech and language therapists (specialist tests of speech and language). All analyses were undertaken using the statistical software package Stata version 14 (Statacorp, College Station, TX, USA).

## RESULTS

One hundred and twenty-five children participated in the full study, of whom 110 (88%) received a diagnosis of CP by age 5 years (Fig S1, online supporting information). Of these, 77 (70%) joined the study at 24 to 35 months of age and were seen again at 5 years (mean 5y 5mo; SD 4mo). No significant difference was observed between the recruits and non-recruits in sex ($\chi^2$ [2 df] 1.85, $p$=0.39) or type of CP ($\chi^2$ [4 df] 5.32, $p$=0.26). Their year of birth differed ($\chi^2$ [8 df] 8.95, $p$=0.02), with fewer children recruited in 2010.

Table 2 shows the characteristics of the 77 participants in the current study. Most children had impairments in all areas of function measured, except hearing. None had ataxic CP. All children with dyskinetic CP had bilateral involvement. We therefore collapsed CP type and distribution into one variable: bilateral spastic, unilateral spastic, or dyskinetic. Six children had congenital malformations of the respiratory system ($n$=3), palate ($n$=1), heart ($n$=1), and eye ($n$=1). One child had a metabolic disorder that did not cause neuromuscular disorder. One data item was missing for six children. Two children lacked non-verbal cognition data and two lacked expressive language data, as assessments were abandoned because they lost concentration during the visit. Data were not recorded for feeding for one child and consonant inventory for another (Table 1). Most children scored at, or below, the first or second centile on standardized tests of language and non-verbal cognition at 2 years. However, there was variation in raw scores for children scoring at the lowest centile rank, so we used raw scores in all analyses. Most children also produced a small range of consonants (mean=2 consonants, mean for typically developing English-speaking children at 24 months=9[31]) and a small number of spoken words (mean=3 words, mean for typically developing children=200[32]). Measures of language and cognition showed strong association at 2 years (Table S1, online supporting information). No differences were observed between children seen at 24 to 29 months of age and those seen at 30 to 35 months of age in levels of impairment, with the exception of GMFCS, and so data were pooled for subsequent analyses. At 5 years of age, three children had a confirmed diagnosis of autism spectrum disorder and three were awaiting diagnosis.

Levels on the CFCS, FCCS, and VSS at 5 years of age showed concordance. CFCS and FCCS levels ($K$=0.78) had the highest correlation (FCCS and VSS [$K$=0.72] and CFCS and VSS [$K$=0.64]) (Table S2, online supporting information).

Univariate regression showed that age at assessment at 2 years, sex, and birthweight were weak predictors of children's CFCS, FCCS, and VSS levels at 5 years. Gestational age predicted CFCS, FCCS, and VSS levels, with later birth associated with more severe levels of impairment. Bilateral spastic CP and dyskinetic CP were associated with more severely impaired communication and speech than unilateral spastic CP. The greatest risk was for children with dyskinesia, especially for VSS level. Hearing was omitted from the univariate analysis because of the low number of children with severe hearing loss. All other areas of function assessed at 2 years were associated with speech and communication classification at 5 years. Severe

**Table 2:** Characteristics of children at 2 years of age

| | Age 24–29mo ($n$=46) | Age 30–35mo ($n$=31) | Total sample ($n$=77) |
|---|---|---|---|
| Age, mo | 26.3 (1.8) | 31.9 (1.7) | 28.6 (3.3) |
| Sex, M:F | 29:17 | 23:8 | 52:25 |
| Gestational age, wks | 35.2 (6.1) | 34.6 (5.1) | 34.9 (5.7) |
| Birthweight ($n$=74), g | 2490.3 (1067.3) 2 missing | 2351.8 (1093.2) 1 missing | 2407.9 (1077.6) 3 missing |
| *CP type, n (%)* | | | |
| Spastic unilateral | 8 (17.4) | 8 (25.8) | 16 (20.8) |
| Spastic bilateral | 23 (50.0) | 16 (51.6) | 39 (50.7) |
| Dyskinetic | 15 (32.6) | 7 (22.6) | 22 (28.6) |
| *Neonatal seizures, n (%)* | | | |
| No | 19 (41.3) | 10 (32.2) | 29 (37.6) |
| Yes | 27 (58.7) | 21 (67.7) | 48 (62.3) |
| *Vision, n (%)* | | | |
| No impairment | 19 (41.3) | 15 (48.4) | 34 (44.2) |
| Impairment but not registered | 15 (32.6) | 11 (35.5) | 26 (33.8) |
| SVI/partially sighted | 12 (26.1) | 16.13) | 17 (22.1) |
| *Hearing, n (%)* | | | |
| No impairment | 43 (93.5) | 30 (96.8) | 73 (94.8) |
| 41–70dB HL | 3 (6.5) | 1 (3.23) | 4 (5.2) |
| *GMFCS level,[a] n (%)* | | | |
| I | 9 (19.6) | 15 (48.4) | 24 (31.2) |
| II | 9 (19.6) | 3 (9.7) | 12 (15.6) |
| III | 3 (6.5) | 3 (9.7) | 6 (7.8) |
| IV | 9 (19.6) | 1 (3.2) | 10 (13.0) |
| V | 16 (34.8) | 9 (29.0) | 25 (32.5) |
| *MACS level, n (%)* | | | |
| I | 6 (13.0) | 2 (6.45) | 8 (10.4) |
| II | 14 (30.4) | 14 (45.16) | 28 (36.4) |
| III | 7 (15.2) | 6 (19.35) | 13 (16.9) |
| IV | 13 (28.3) | 4 (12.9) | 17 (22.1) |
| V | 6 (13.0) | 5 (16.13) | 11 (14.3) |
| *Feeding method (n = 76), n (%)* | | | |
| Oral no problem | 8 (17.4) | 4 (12.9) | 12 (15.6) |
| Oral with oral motor problem | 34 (73.9) | 18 (58.0) | 52 (67.5) |
| Oral with tube/tube | 4 (8.7) | 9 (29.0) | 13 (16.9) |
| *Non-verbal cognition (n = 75)* | | | |
| Raw score | 18.5 (9.3) | 21.9 (9.8) | 19.8 (9.6) |
| Centile rank, median (IQR) | 1 (1–15) | 1 (1–31) | 1 (1–21) |
| *Receptive language* | | | |
| Raw score | 17. 4 (8.2) | 22.6 (14.7) | 19.5 (11.5) |
| Centile rank, median (IQR) | 1 (1–12) | 1 (1–6) | 2 (1–11) |
| *Expressive language (n = 75)* | | | |
| Raw score | 15.3 (9.2) | 20.8 (16.0) | 17.6 (12.7) |
| Centile rank, median (IQR) | 2 (1–5.5) | 1 (1–5) | 1 (1–5) |
| MCDI words, median (IQR) | 3 (0–59) | 4 (0–133) | 3 (0–83) |
| Number of consonants ($n$=76), median (IQR) | 2 (0–8) | 2 (0–9) | 2 (0–9) |
| *Speech understood, n (%)* | | | |
| All/almost all | 5 (10.9) | 2 (6.5) | 7 (9.1) |
| Three-quarters | 3 (6.5) | 3 (9.7) | 6 (7.8) |
| One-half | 8 (17.4) | 8 (25.8) | 16 (20.8) |
| Less than one-half | 30 (65.2) | 18 (58.1) | 48 (62.3) |

Data are mean (SD) unless otherwise stated. [a]Gross Motor Function Classification System (GMFCS) level statistically significant difference between children aged 24 to 29 months and 30 to 35 months ($\chi^2$ [df 4] 10.3, $p$=0.03). SD, standard deviation; M, male; F, female; CP, cerebral palsy; SVI, severe visual impairment; HL, hearing loss; MACS, Manual Ability Classification System; IQR, interquartile range; MCDI, MacArthur Communicative Development Inventory.

**Table 3:** Univariable prediction of CFCS, FCCS, and VSS outcomes at 5 years of age from characteristics at 2 years of age

| | CFCS at 5 years | | FCCS at 5 years | | VSS at 5 years | |
|---|---|---|---|---|---|---|
| | OR | 95% CI | OR | 95% CI | OR | 95% CI |
| Age, mo | 0.99 | 0.87–1.11 | 1.00 | 0.88–1.14 | 0.96 | 0.84–1.09 |
| Sex (reference group=male) | 1.8 | 0.76–4.27 | 1.45 | 0.63–3.38 | 1.25 | 0.53–2.97 |
| Gestational age (wks) | 1.08 | 1.00–1.17 | 1.09 | 1.01–1.74 | 1.09 | 1.01–1.18 |
| Birthweight (g) | 1.00 | 1.00–1.00 | 1.00 | 1.00–1.00 | 1.00 | 1.00–1.00 |
| *CP type* | | | | | | |
| Spastic unilateral | 1 | — | 1 | — | 1 | — |
| Spastic bilateral | 9.60 | 3.00–30.75 | 9.99 | 3.02–33.11 | 7.28 | 2.30–23.05 |
| Dyskinetic | 12.19 | 3.52–42.19 | 12.55 | 3.51–44.90 | 18.11 | 4.81–68.12 |
| *Neonatal seizures* | | | | | | |
| No | 1 | — | 1 | — | 1 | — |
| Yes | 5.08 | 2.10–12.33 | 3.66 | 1.56–8.62 | 4.11 | 1.68–10.03 |
| *Vision* | | | | | | |
| No impairment | 1 | — | 1 | — | 1 | — |
| Impairment, not registered | 2.20 | 0.87–5.58 | 1.04 | 0.80–5.21 | 1.22 | 0.49–3.01 |
| SVI/partially sighted | 35.02 | 8.51–114.19 | 31.56 | 7.64–130.5 | 7.17 | 1.94–26.48 |
| *GMFCS level* | | | | | | |
| I | 1 | — | 1 | — | 1 | — |
| II | 0.51 | 0.15–1.76 | 0.69 | 0.19–2.50 | 0.89 | 0.25–3.11 |
| III | 1.49 | 0.29–7.67 | 2.95 | 0.57–15.16 | 2.69 | 0.54–13.47 |
| IV | 2.41 | 0.60–9.62 | 2.84 | 0.67–11.97 | 4.82 | 1.04–22.19 |
| V | 92.07 | 16.62–510.0 | 45.02 | 11.14–182.0 | 51.08 | 12.22–213.45 |
| *MACS level* | | | | | | |
| I | 1 | — | 1 | — | 1 | — |
| II | 1.26 | 0.29–5.53 | 1.81 | 0.39–8.36 | 1.29 | 0.30–5.54 |
| III | 1.51 | 0.29–7.71 | 2.05 | 0.38–11.02 | 1.70 | 0.34–8.52 |
| IV | 41.07 | 6.12–275.76 | 37.26 | 5.81–239.10 | 43.70 | 7.01–272.40 |
| V | 521.49 | 45.45–5984 | 404.16 | 40.05–4078 | $5.25^{08}$ | 0 |
| *Feeding method* | | | | | | |
| Oral no problem | 1 | — | 1 | — | 1 | — |
| Oral with difficulty | 2.28 | 0.73–7.15 | 2.97 | 0.93–9.45 | 5.91 | 1.77–19.74 |
| Oral with tube/tube | 11.12 | 2.35–52.52 | 11.99 | 2.56–56.16 | 18.34 | 3.56–94.37 |
| Non-verbal cognition, *rs* | 0.78 | 0.72–0.84 | 0.79 | 0.74–0.85 | 0.83 | 0.78–0.89 |
| Receptive language, *rs* | 0.90 | 0.85–0.95 | 0.89 | 0.85–0.94 | 0.91 | 0.86–0.96 |
| Expressive language, *rs* | 0.89 | 0.85–0.93 | 0.89 | 0.84–0.93 | 0.89 | 0.85–0.93 |
| MCDI words | 0.99 | 0.98–0.99 | 0.99 | 0.99–0.99 | 0.99 | 0.99–0.99 |
| Number of consonants | 0.65 | 0.57–0.74 | 0.65 | 0.56–0.74 | 0.65 | 0.57–0.75 |
| *Speech understood* | | | | | | |
| All/almost all | 1 | — | 1 | — | 1 | — |
| Three-quarters | 5.01 | 0.73–34.28 | 2.71 | 0.38–19.10 | 3.04 | 0.43–21.48 |
| One-half | 7.56 | 1.19–48.06 | 5.95 | 0.88–39.98 | 6.90 | 1.03–46.09 |
| Less than one-half | 596.36 | 63.27–5621.6 | 269.35 | 31.57–2298.0 | 363.41 | 41.05–3217.3 |

CFCS, Communication Function Classification System; FCCS, Functional Communication Classification System; VSS, Viking Speech Scale; OR, odds ratio; CI, confidence interval; CP, cerebral palsy; SVI, severe visual impairment; HL, hearing loss; GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System; *rs*, raw score; MCDI, MacArthur Communicative Development Inventory.

visual impairment was a stronger predictor of communication (CFCS and FCCS) than speech (VSS). MACS appeared to be a stronger predictor of speech and communication than GMFCS. Only GMFCS level V was associated with a different level on the CFCS, FCCS, or VSS, whereas MACS levels IV and V were both significant predictors of speech and communication levels (Table 3).

Because of the lack of an age association, we did not adjust multivariable logistic regression models for age. Given the lack of difference between levels I to III in the univariate regression models, we reclassified MACS levels I to III, forming a three-level categorical variable—I to III, IV, and V—for the multivariable analysis. We retained all levels of the GMFCS because it previously predicted communication outcome in its full form.[10]

Multivariable models using data available to paediatricians indicated that CP type and distribution, GMFCS, vision, and amount of speech understood by strangers predicted the CFCS level ($R^2$=0.51) (Table 4). The fit was similar for speech and language therapy (SLT) data when non-verbal cognition raw score at 2 years was added to the model and vision was removed ($R^2$=0.52).

For the FCCS, the best fit model using measures available to paediatricians included CP type and distribution, the collapsed MACS level, the amount of speech understood by strangers, and vision ($R^2$=0.43). For SLT data, the model improved slightly when the number of consonants was added ($R^2$=0.49).

The best fit model to predict VSS level using data available to paediatricians included CP type and distribution and amount of speech understood by strangers only ($R^2$=0.34). The best fit model for SLT data available included CP type, speech understood by strangers, and number of consonants ($R^2$=0.50).

## DISCUSSION

This study showed that for children whose communication is causing concern at 2 years of age, demographic characteristics, early medical history, type and distribution of CP, and functional performance measures predict the severity of speech and communication difficulty at 5 years. Data available to paediatricians at 2 years can predict speech and communication at 5 years, but models slightly improve with the addition of specialist speech, language, and cognition measures. Although strengths of regression analyses were similar, the variables that predict CFCS, FCCS, and VSS levels differ, supporting the divergent validity of the three classification systems.

As in previous research, we found that the risk of speech and/or communication difficulties was greater for children with dyskinetic than spastic forms of CP,[3,5,7] for children born at term, those with slightly higher birthweight,[1] those who had neonatal seizures,[1] and for children with more severely impaired vison,[1,12] gross and fine motor function, feeding impairment,[1] and non-verbal cognition.[3,5,9–11]

The measures that continued to predict CFCS levels in our multivariable models—CP type and distribution, gross motor function (GMFCS level V), and speech (amount of speech understood by strangers)—were similar to the two previous studies of the CFCS.[2,12] However, unlike the previous studies, vision (severe visual impairment) and non-verbal cognition remained significant predictors of CFCS level in our multivariable models after adjustment for other variables. Differences may be because of sampling; the previous studies included any child with CP, while ours focused on children who had communication concerns at 2 years of age. Unlike this study, Hidecker et al.[12] observed prediction of CFCS level by gestational age in multivariable models. We measured gestational age in completed weeks, while Hidecker et al. used a binary measure of birth on or before 32 completed weeks or later. Approximately one-third of our sample had dyskinesia, which is associated with term delivery. Our lack of continued prediction by gestational age could be caused by association with motor disorder type or a lack of statistical power. Further research, containing a spread of characteristics and powered to detect differences within groups, is required to refine our estimates of CFCS prediction.

To our knowledge, this is the first study of the prediction of FCCS or VSS levels. CP type and distribution, MACS, vision, amount of speech understood by strangers, and number of consonants spoken at 2 years of age were the best predictors of FCCS level. All factors reflect the physical and cognitive aspects involved in observable expressive communication that the FCCS aims to classify, and that can be accomplished verbally, through gesture, or by using AAC. Previous research has shown a relationship between cognition and MACS level,[33] which could explain why cognition dropped out of this multivariable model. The fact that cognition was retained in the multivariable model for CFCS could relate to the rating of comprehension within the CFCS in both the sending and receiving of messages, as cognition correlated highly with receptive language measures in our sample.

Only CP type and distribution, amount of speech understood, and number of consonants predicted VSS levels. Hustad et al. also noted that speech development and spoken language at 2 years predicted speech and language function at 5 years.[14,15] The small set of predictor variables for the VSS, compared to those for the CFCS and FCCS communication levels, provide additional support for the construct validity of the VSS as a measure of speech only. Similarly, the retention of vision and motor function in multivariable models of CFCS and FCCS prediction highlight the role of non-verbal expression in communication.

All multivariable models retained CP type and distribution as a predictor of communication and speech outcome. Moreover, in all but one model (the FCCS using paediatrician data), a clear order of increasing risk of communication difficulty occurred for children with unilateral spastic, bilateral spastic, and dyskinetic forms of CP. Future research should therefore consider the communication profiles of children with these different types and distributions of motor disorders separately.

The multivariable models accounted for approximately one-half of the variance in the CFCS, FCSS, and VSS. We cannot compare this with previous research, as variance was not reported previously.[2,10,13] The remaining 50% is likely to represent inter- and intra-child variability. Communication always arises from a desire to express a message, involves two partners, is regulated by sociocultural norms, and is affected by the physical environment such as background noise. Future investigations should include measures of personal and environmental factors to examine their influence on communication development. Reporting of the amount of variance explained by regression models would help build understanding of the interrelationships between predictive factors and could inform interventions.

### Implications

The predictions of CFCS, FCCS, and VSS levels in our multivariable models using data readily available to paediatricians can be used to inform discussion of risk estimates for communication difficulties in early childhood consultations. Children with bilateral spastic or dyskinetic motor disorder, severe visual impairment, or no intelligible speech sounds or less than half their speech is understood at 2 years of age should be referred to SLT. They are at the most severe risk of severe speech and communication difficulties, and may require AAC.

SLT assessment at 2 years should include non-verbal cognition and speech sound system assessment, as these early skills predict communication at 5 years and could guide intervention, including decisions around the most suitable type of AAC to supplement or replace speech. Further research, with larger samples including the measures tested here, is needed to create profiles of impairment severity (CFCS, FCCS, and VSS levels). The first step could be the amalgamation of existing international data sets.

**Table 4:** Multivariable prediction of CFCS, FCCS, and VSS outcomes at 5 years of age, by characteristics and measures available to paediatricians, and speech and language therapists, at 2 years of age

| | CFCS at 5 years | | | | FCCS at 5 years | | | | VSS at 5 years | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | Paediatrician | | Speech and language therapist | | Paediatrician | | Speech and language therapist | | Paediatrician | | Speech and language therapist | |
| | OR | 95% CI | OR | 95% CI | OR | 95% CI | OR | 95% CI | OR | 95% CI | OR | 95% CI |
| *Type CP* | | | | | | | | | | | | |
| Spastic unilateral | 1 | — | 1 | — | 1 | — | 1 | — | 1 | — | 1 | — |
| Spastic bilateral | 9.33 | 2.08–41.75 | 19.18 | 3.40–108.3 | 8.44 | 1.99–35.65 | 6.35 | 1.36–29.66 | 9.12 | 2.43–34.26 | 4.73 | 1.00–22.38 |
| Dyskinetic | 9.10 | 1.86–44.41 | 33.92 | 4.88–235.6 | 7.63 | 1.61–36.33 | 8.59 | 1.62–45.43 | 17.41 | 3.88–78.17 | 29.34 | 4.61–186.7 |
| *Vision* | | | | | | | | | | | | |
| No impairment | 1 | — | | | 1 | — | 1 | — | | | | |
| Impaired, not reg. | 2.21 | 0.75–6.60 | | | 2.14 | 0.71–6.44 | 1.88 | 0.58–6.16 | | | | |
| SVI/partially sighted | 30.39 | 5.80–159.4 | | | 29.12 | 4.82–175.8 | 20.39 | 3.15–131.8 | | | | |
| *GMFCS* | | | | | | | | | | | | |
| I | 1 | — | 1 | — | | | | | | | | |
| II | 0.15 | 0.03–0.72 | 0.09 | 0.01–0.53 | | | | | | | | |
| III | 0.45 | 0.07–2.85 | 0.06 | 0.01–0.58 | | | | | | | | |
| IV | 0.79 | 0.16–3.93 | 0.11 | 0.01–0.89 | | | | | | | | |
| V | 10.10 | 1.43–71.11 | 2.31 | 0.19–26.98 | | | | | | | | |
| *MACS* | | | | | | | | | | | | |
| I–III | | | | | 1 | — | 1 | — | | | | |
| IV | | | | | 2.67 | 0.66–12.53 | 2.46 | 0.56–10.90 | | | | |
| V | | | | | 21.16 | 2.18–204.9 | 14.97 | 1.61–138.8 | | | | |
| Non-verbal cognition *rs* | | | 0.81 | 0.72–0.90 | | | | | | | | |
| Number of consonants | | | | | | | 0.75 | 0.63–0.88 | | | 0.63 | 0.51–0.77 |
| *Speech understood* | | | | | | | | | | | | |
| All/almost all | 1 | — | 1 | — | 1 | — | 1 | — | 1 | — | 1 | — |
| Three-quarters | 1.90 | 0.16–23.14 | 6.57 | 0.65–66.98 | 1.88 | 0.15–22.80 | 7.82 | 0.36–169.4 | 2.48 | 0.22–28.53 | 41.92 | 1.3–1352.3 |
| One-half | 5.67 | 0.70–46.10 | 3.82 | 0.41–35.87 | 7.71 | 0.93–63.56 | 19.62 | 1.63–236.8 | 6.28 | 0.78–50.43 | 48.99 | 2.75–871.5 |
| Less than one-half | 43.35 | 5.50–342.3 | 214.2 | 11.97–3833 | 106.5 | 11.30–10004.9 | 71.67 | 5.28–971.6 | 168.9 | 17.87–1595.8 | 120.2 | 6.74–2144.5 |

CFCS, Communication Function Classification System; FCCS, Functional Communication Classification System; VSS, Viking Speech Scale; OR, odds ratio; CI, confidence interval; CP, cerebral palsy; SVI, severe visual impairment; GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System; *rs*, raw score.

Previous research has suggested that different constructs are measured by the CFCS, FCCS, and VSS, and recommended that CP registers include the VSS and either the CFCS or FCCS. Our findings support the divergent validity of the three rating scales and that all the current classification schemes—the GMFCS, MACS, Eating and Drinking Ability Classification System, CFCS, FCCS, and VSS—provide different but complementary information with which to profile children's function. We would recommend that all are used in future research and should form part of the common data set for CP.[34]

Similar to Mei et al.,[11] we found that measures of receptive and expressive language and non-verbal cognition were strongly associated. This included the four-point speech intelligibility scale validated by Coplan and Gleason,[23] which to our knowledge has not been used with children with CP previously. As the scale predicted all outcomes and was easy to complete by parents,[23] it should be added to clinical investigations at 2 years of age.

## Strengths and limitations

This is the largest prospective sample of children with CP and communication difficulties reported to date, and validated measures of speech, language, and cognition were undertaken by trained therapists. Children with severe motor disorders were assessed using standardized assessments of receptive language and non-verbal cognition, some with adaptations to response. For some children, adaptations will have increased the cognitive processing required and results may underestimate their true abilities.[35] Receptive language tests for children with little movement are being developed and should address this issue in future research.[36] This is a novel cohort, specifically recruited to assess which domains best predict speech and communication for children whose communication was already causing concern at 2 years of age. Our aim necessitated testing all domains that had previously been associated with speech and communication in population studies of CP, resulting in multiple testing that introduces the risk of a type I error. Ours was a convenience sample and, while not different from the population from which it was recruited in terms of sex, or type or distribution of CP, it may not represent the full range of children with CP whose speech, language, or communication is delayed or different to their typically developing peers. Although some children's results on measures of speech, language, or cognition were within the range expected for children of their age, many children in our sample had severe difficulties at 2 years. The study may have been more attractive to families of these children as a source of information gathering. We were not able to assess if the children who were identified but not recruited to the study had less-severe communication difficulties. The resulting small subgroups also limited the precision of our predictions.

## CONCLUSION

Communication and speech performance at 5 years of age are best predicted by CP type and distribution, and spoken language, as rated by the four-point scale by Coplan and Gleason,[23] at 2 years. Motor function, cognition, and vision are also important predictors of communication, but not speech. Children with bilateral spastic or dyskinetic CP, and little spoken language by 2 years, are at the highest risk of speech and communication difficulties at 5 years of age, and should be referred to SLT for advice on implementing AAC to promote communication development and facilitate social participation.


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## SUPPORTING INFORMATION

The following additional material may be found online:

**Table S1:** Correlations between measures of language and non-verbal cognition at 2 years of age.

**Table S2:** Associations between CFCS, FCCS, and VSS at 5 years of age.

**Figure S1:** Participant flow.

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## TRASTORNOS DE LA COMUNICACIÓN EN NIÑOS PEQUEÑOS CON PARÁLISIS CEREBRAL


### OBJETIVO
Probar la predicción de la gravedad del trastorno de comunicación a los 5 años a partir de las características a los 2 años para niños con parálisis cerebral (PC) cuya comunicación es motivo de preocupación.

### MÉTODO
En este estudio de cohorte, 77 niños (52 varones; 25 mujeres) con dificultades de comunicación y PC fueron visitados en el hogar a los 2 años (media 2 años y 4 meses; DE 3 meses) y 5 (media 5 años y 5 meses; DE 4 meses). La información sobre el tipo y la distribución del trastorno motor, las convulsiones, la función motora gruesa y fina, la audición y la visión se obtuvieron de las notas médicas. La cognición no verbal, la comprensión del lenguaje, la expresión del lenguaje, el vocabulario hablado y los métodos de comunicación se evaluaron directamente a los 2 años. A los 5 años, la función de comunicación y habla se calificó utilizando el Sistema de Clasificación de Función de Comunicación (CFCS), el Sistema de Clasificación de Comunicación Funcional (FCCS) y la Escala de Habla Vikinga.

### RESULTADOS
En los modelos de regresión multivariable, el tipo de PC, el nivel del sistema de clasificación de la función motora gruesa, la visión, la cantidad de lenguaje que entienden los extraños, la cognición no verbal y el número de consonantes producidas a los 2 años predijeron el nivel de CFCS a los 5 años (R2 = 0,54). El tipo de PC, el nivel del Sistema de Clasificación de Habilidad Manual, la cantidad de discurso entendido, la visión y el número de consonantes predijeron el nivel de FCCS (R2 = 0,49). El tipo de PC, la cantidad de discurso que entienden los extraños y el número de consonantes predijeron el nivel de la Escala de discurso vikingo (R2 = 0,50).

### INTERPRETACIÓN
Las características a los 2 años predicen la comunicación y el rendimiento del habla a los 5 años, y deben informar la derivación a la consulta de terapia del habla y el lenguaje.


## TRANSTORNOS DE COMUNICAÇÃO EM CRIANÇAS PEQUENAS COM PARALISIA CEREBRAL


### OBJETIVO
Testar a predição da severidade de transtornos de comunicação aos 5 anos de idade a partir de características aos 2 anos para crianças com paralisia cerebral (PC) cuja comunicação está causando preocupação.

### MÉTODO
Nestes estudo de coorte, 77 crianças (52 do sexo masculino; 25 do sexo feminino) com dificuldades de comunicação e PC foram visitadas em casa aos2 (média 2a 4m; DP 3m) e 5 (média 5a 5m; DP 4m) anos de idade. Informação sobre o tipo e distribuição da desordem motora, convulsões, função motora grossa e fina, audição e visão foram coletados em registros médicos. Cognição não verbal, compreensão da linguagem, expressão da linguagem, vocabulário falado, e métodos de comunicação foram avaliados diretamente aos 2 anos. Aos 5 anos, a função de comunicação e fala foram pontuadas usando o Sistema de Classificação da Função de Comunicação (SCFC), Sistema de Classificação da Comunicação Funcional (SCFC) e Escala Viking de Fala.

### RESULTADOS
Em modelos multivariados de regressão, o tipo de PC, nível do Sistema de Classificação da Função Motora Grossa, visão, quantidade de fala compreendida por estranhos, cognição não verbal, e número de consoantes produzidas aos 2 anos de idade predizeram o nível SCFC aos 5 anos de idade ($R^2$=0,4). O tipo de PC, nível do Sistema de Classificação da Função Manual, quantidade de fala compreendida por estranhos, e número de consoantes produzidas aos 2 anos de idade predizeram o nível eSCFC ($R^2$=0,49). O tipo de PC, nível do Sistema de quantidade de fala compreendida por estranhos, e número de consoantes predizeram o nível da Escala Viking de Fala Speech Scale level ($R^2$=0,50).

### INTERPRETAÇÃO
Características aos 2 anos de idade foram preditivas do desempenho em comunicação e fala aos 5 anos, e devem informar o encaminhamento para terapias de fala e linguagem.