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Title:

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Date:

2020-04-01

Citation:

Sanchez, K., Boyce, J. O., Mei, C., St John, M., Smith, J., Leembruggen, L., Mills, S., Spittle, A. J. & Morgan, A. T. (2020). Communication in children born very preterm: a prospective cohort study. *Developmental Medicine and Child Neurology*, 62 (4), pp.506-512. <https://doi.org/10.1111/dmcn.14360>.

Persistent Link:

<https://hdl.handle.net/11343/286432>

Author Manuscript

This is the author manuscript accepted for publication and has undergone full peer review but has not been through the copyediting, typesetting, pagination and proofreading process, which may lead to differences between this version and the [Version of Record](#). Please cite this article as [doi: 10.1111/dmcn.14360](https://doi.org/10.1111/dmcn.14360)

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Article type : Original Article

[Original article: 3 tables; 1 figure; 2 online tables; 1 online figure]

Communication in children born very preterm: a prospective cohort study

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PUBLICATION DATA

Accepted for publication 00th Month 2019.

Published online 00th Month 2019.

ABBREVIATIONS

DEAP	Diagnostic Evaluation of Articulation and Phonology
GRBAS	Grade Roughness Breathiness Asthenia Strain
PLS-5	Preschool Language Scales, Fifth Edition
pVHI	Pediatric Voice Handicap Index

[Abstract]

AIM To compare language, speech, and voice of children born preterm and at term, and determine relevant predictors of outcome.

METHOD Three hundred infants (150 males, 150 females; 149 born at <30wks' gestation, 151 term-born) were prospectively recruited at birth from the Royal Women's Hospital. We administered the Preschool Language Scales, Fifth Edition, Diagnostic Evaluation of Articulation and Phonology, Grade Roughness Breathiness Asthenia Strain Scale, and Pediatric Voice Handicap Index at 3 years, and compared groups. We examined hypothesized predictors in children born preterm: gestational age at birth, birthweight, sex, chronic lung disease, high social risk, multilingualism, neurodevelopmental diagnosis, and oromotor feeding.

RESULTS Children born preterm had poorer language than children born at term (coefficient -5.43). Speech and voice were similar between groups (coefficients -0.70 to 1.63). Chronic lung disease predicted voice (coefficient 6.05); male sex (coefficients 4.54–6.18), high social risk (coefficient -6.02 to -9.30), and neurodevelopmental diagnosis (coefficients -16.42 to -20.61) predicted language.

INTERPRETATION Children born before 30 weeks' gestation had poorer language than children born at term. Children born preterm with neurodevelopmental disabilities or high social risk experience poorer language outcomes, and would benefit from enrichment of their language environment.

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DOI: 10.1111/dmcn.xxxxx

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Developmental Medicine & Child Neurology 2019, 61: 000–000

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Communication in Children Born Very Preterm Katherine Sanchez et al.

What this paper adds

- Speech and voice outcomes were similar between children born preterm and at term.
- Male sex, high social risk, and neurodevelopmental diagnosis predicted language outcomes.

[Main text]

Developmental speech and language impairments have a deleterious effect on the quality of life of children and their parents,¹ and affect literacy, educational, and employment outcomes.² Children born preterm have a heightened risk of communication (language, speech, and voice) difficulties. They consistently present with poorer language than their term-born peers,³ and may also experience difficulties with voice and speech sound production.^{4,5} However, to date, research has not reflected a complete picture of communication ability in children born preterm. Reports have typically focused on isolated domains of communication such as receptive vocabulary.³ This restricts knowledge of the overall presentation and significant risk factors contributing to language, speech, and voice outcomes, which limits capacity for targeting and planning intervention.

Effective communication requires competence across language, speech, and voice. Language difficulties include deficits in receptive (understanding) and expressive (production) domains, across the areas of phonology, syntax, word finding and semantics, pragmatics, discourse, and verbal learning and memory, and may occur in isolation or as part of a broader profile of functional difficulties.⁶ Speech difficulties are concerned with speech sound production, and include linguistically based speech problems (such as phonological errors, e.g. pronouncing all fricative sounds like ‘s’ and ‘f’ as stopped sounds like ‘t’ and ‘p’) and problems with the physical production of sounds (such as articulation errors, e.g. isolated lisps).⁷ Voice difficulties comprise problems with resonance, phonation, and respiration for speech, for example a voice that sounds hoarse or breathy. Language, speech, and voice

difficulties are known to co-occur in many children,⁸ perhaps compounding the known effects on quality of life.

The exploration of language, speech, and voice development in the preterm population has been hindered by a number of factors. Most research has assessed language abilities using screening measures (e.g. Bayley Scales of Infant and Toddler Development) or unidimensional measures (e.g. expressive vocabulary subscale of the Wechsler Intelligence Scale for Children), which are not designed to comprehensively explore or reliably diagnose language difficulties.³ Speech abilities have been underexamined in children born preterm. Many studies of speech use brief screeners, informal observation, or unvalidated tools, and frequently make no distinction between clinically significant phonological versus articulatory or other motor speech difficulties.⁵ This distinction is critical as phonological and articulatory speech difficulties require different therapeutic approaches. One study used rigorous assessment methods that distinguished between types of speech difficulties, and found some evidence of deficits in speech motor control in children born preterm.⁹ Similar research with younger children born preterm is lacking, which has significant implications for the early identification and remediation of speech sound difficulties. Likewise, voice has been infrequently studied among children born preterm, with a 2014 review identifying the need for further research with a particular focus on significant risk factors.⁴ Current studies of voice are indicated, especially given the evolution of respiratory management in infants born preterm, and the fact that intubation has been highlighted as a predictor of voice outcomes.^{4,10}

Given that children born preterm appear to have a heightened risk of language, speech, and voice difficulties, establishing relevant predictors is paramount. Knowledge of significant risk factors can inform a differential risk model to assist in targeting surveillance and direct intervention services towards children at greatest need. A diverse range of significant risk factors have been suggested, from population-level risk factors such as sex and social risk¹¹ to factors more specific to preterm birth such as gestational age at birth,¹² birthweight,¹² chronic lung disease,¹³ and oromotor deficits (often measured via feeding skills).⁹

In this prospective cohort study, our core aims were to: (1) explore the differences between language, speech, and voice of children born preterm and at term at 3 years' corrected age; and (2) explore predictors of language, speech, and voice outcomes in children born preterm at 3 years of age. A further aim was to report co-occurrence of language, speech, and voice difficulties within the preterm population.

METHOD

Participants

Three hundred infants were recruited at birth from the Royal Women's Hospital and Frances Perry House Private Hospital, Melbourne, between January 2011 and March 2013.¹⁴ Both hospitals allow parental presence at any time. Infants with congenital abnormalities known to affect neurodevelopment or those with non-English-speaking parents were excluded.¹⁴ We determined the cohort size from the number of infants aged less than 30 weeks cared for each year at Royal Women's Hospital, and the expected consent rate for the study. The Royal Women's Hospital Human Research Ethics Committee (10/07) and Royal Children's Hospital Research Ethics Committee (34147A) approved this study and caregivers provided written consent.

Collection of predictor data

Four registered research nurses and the first author (KS) collected demographic and medical data at birth via parent questionnaires and medical chart review, including sex, gestational age at birth, birthweight z-score,¹⁵ chronic lung disease, and social risk (using the Social Risk Index, a score out of six that evaluates six aspects of social status: family structure; education of primary caregiver; occupation of primary income earner; employment status of primary income earner; language spoken at home; and maternal age at birth).¹⁴ The recruitment sites were located in a region with universal hearing screening, suggesting that significant undetected congenital sensorineural hearing impairments were unlikely. Oromotor feeding at 12 months was evaluated using a validated assessment tool, and children were assigned to a delayed or typical group, depending on their performance, to facilitate predictor analysis (detailed methods and results of this assessment are provided in an earlier paper).¹⁶

Three-year data collection

When their child was approximately 36 months' corrected age, parents were asked to complete the Pediatric Voice Handicap Index (pVHI)¹⁷ electronically using REDCap,¹⁸ and complete a custom questionnaire that asked about home languages; pre-existing diagnoses of neurodevelopmental disorders, including cerebral palsy, autism spectrum disorder, or global developmental delay; whether there had been any parent or professional concerns about language, speech, or voice; and whether the child had ever attended speech pathology for these areas of development. Families were also invited to attend a face-to-face assessment where the child completed a range of standardized assessments administered by one of five

trained paediatric speech pathologists (KS, JB, CM, MSJ, JS). The assessment protocol included the full Preschool Language Scales, Fifth Edition (PLS-5 - Australia and New Zealand Adapted);¹⁹ the Articulation, Phonology and Oro-Motor subtests of the Diagnostic Evaluation of Articulation and Phonology (DEAP);²⁰ and the Grade Roughness Breathiness Asthenia Strain (GRBAS) voice-rating scale.²¹ The DEAP and GRBAS assessment samples were audio-recorded for interrater reliability purposes.

Variables of interest for this study are listed below.

Speech

Speech sound difficulties were assessed based on the norms for acquisition of specific sounds and elimination of phonological processes from the DEAP Articulation and Phonology subtests.²⁰ Continuous variables used for predictor analyses were: percentage of consonants correct standard score, number of consonants produced (including stimulability – a measure of articulation), and number of phonological processes (a measure of phonology). The number of children scoring more than one standard deviation (SD) below the mean on the DEAP Oro-Motor subtest was also reported.

Voice

Voice difficulties were based on a GRBAS grade score of 2 or more and/or 1.5 SDs above term mean pVHI total score. Continuous variables used for predictor analyses were GRBAS grade score and total pVHI score.

Language

Language difficulties were based on a standard score less than 85 on any of the three PLS-5 indices. Continuous variables used for predictor analyses were the auditory comprehension, expressive communication, and total language scores from the PLS-5.

Interrater reliability was evaluated for the DEAP and GRBAS using audio- and video-recordings. A paediatric speech pathologist (JB) rescored 22 randomly selected assessments, comprising 11 term and 11 preterm samples, none of which had been originally scored by JB. Fleiss's Kappa was calculated for binary data and the intraclass correlation coefficient for continuous data. Values of over 0.75 were considered excellent, between 0.40 and 0.75 were fair to good, and less than 0.40 were poor.²²

Statistical analysis

Data were entered into REDCap¹⁸ and analysed using Stata 14 statistical software (StataCorp, College Station, TX, USA). Between-group analyses involving the GRBAS scale were controlled for the presence of upper respiratory tract infections on the day of assessment. Group differences (aim 1) and potential predictors (aim 2) of language, speech, and voice outcomes were evaluated via linear regression fitted using generalized estimating equations to account for multiple births or ordered logistic regression clustered for multiple births. Three continuous variables (number of consonants produced, number of phonological processes, and pVHI total score) were not normally distributed. For one of these variables (pVHI total score) a logarithmic transformation was applied before analysis, which resulted in normal distribution. Regarding the other two variables, logarithmic transformation did not result in normal distribution, and so robust standard errors were used to compensate for non-normality.

In analysing potential predictors, univariable analysis was conducted first for all predictors, with only significant predictors in the univariable analysis then added to the multivariable analysis. A significance level of $p < 0.05$ was adopted. No adjustments for multiple comparisons were made.

Hypothesized predictors were selected on the basis of having been significantly predictive of language, speech, or voice outcomes in previous studies. Sensorineural hearing loss was a planned predictor for analysis but was omitted owing to low numbers ($n=2$). Predictors of interest included sex, gestational age at birth, birthweight z-score, chronic lung disease (oxygen at 36wks), high social risk (score ≥ 2 on Social Risk Index), multilingualism, neurodevelopmental diagnosis (as reported by parents), and oromotor feeding impairment at 12 months (defined as failing one or more food challenges on the Schedule for Oral Motor Assessment).¹⁶

RESULTS

Participants

The follow-up rate for the 3-year assessment was 74% (78% of surviving children born at <30 wks' gestation, 70% of children born at term). Follow-up rates were determined based on completion of any component of the follow-up assessment. The recruitment and retention of participants over the 3 years of the study are summarized in the Figure S1 (online supporting information). Table 1 reports the characteristics of the children who participated in the study.

Sample characteristics were similar between the entire group recruited at birth and those who participated in the 3-year follow-up assessment.

Interrater reliability

Interrater reliability values for the DEAP and GRBAS are reported in Table S1 (online supporting information). Reliability was excellent for the number of consonants produced and number of phonological processes, moderate for the percentage of consonants correct standard score, and poor for the GRBAS grade score.²² Percentage agreement was over 90% for both diagnosis of speech sound difficulties, and perceptual voice difficulties (GRBAS Grade score $\geq$ moderate).

Language, speech, and voice outcomes

Speech sound difficulties were present in 32.7% of the preterm group, and 29.2% of the term group. Of the children with speech sound difficulties, 29 (16 preterm, 13 term) had both articulation and phonological difficulties; nine (four preterm, five term) had phonological difficulties but no articulation difficulties; and 20 (10 preterm, 10 term) had articulation difficulties but no phonological difficulties. Two participants (both preterm) had articulation difficulties but did not complete the phonological test; and one (preterm) had phonological difficulties but did not complete the articulation test.

Voice difficulties were present in 17.2% of the preterm group and 10.1% of the term group.

Language difficulties were present in 19.6% of the preterm group and 4.3% of the term group. Of the children with language difficulties, five (four preterm, one term) had low scores (<85) on all three indices of the PLS-5; one (preterm) scored low only on Total Language; six (four preterm, two term) scored low only on Auditory Comprehension; six (five preterm, one term) scored low on Auditory Comprehension and Total Language but within typical range for Expressive Communication; and one (preterm) scored low for Expressive Communication and Total Language but within typical range for Auditory Comprehension. No child scored low only on Expressive Communication; or scored within typical range for Auditory Comprehension and Expressive Communication but low for Total Language.

Figure 1 demonstrates the overlap between language, speech, and voice difficulties for those children in the preterm group who completed all assessments. It shows that most children born preterm presented with no language, speech, or voice difficulties ($n=37$),

followed by a single area of difficulty (n=29), while a relatively small number demonstrated overlapping areas of difficulty (n=10). An oromotor impairment was present in 6.8% of the preterm group and 0% of the term group.

In Table 2, continuous speech, voice, and language outcomes from the DEAP, GRBAS, and PLS-5 are presented. The preterm group scored substantially lower than the term group on auditory comprehension, expressive communication, and total language, but there were no discernible differences between the groups on speech or voice outcomes.

Predictors of language, speech, and voice outcomes

The results of univariable analysis of predictors are available in Table S2 (online supporting information). Speech intelligibility (percentage of consonants correct standard score), phonology (number of phonological processes), and perceptual voice score (GRBAS grade score) were not associated with any of the examined risk factors. All other outcomes had multiple associations on univariable analysis; therefore, subsequent multivariable analysis was conducted to determine independent predictors (Table 3). Poorer voice functioning (pVHI total score) was predicted by chronic lung disease (coefficient 6.05, 95% confidence interval [CI] 1.21–10.89; $p=0.02$); auditory comprehension (PLS-5) by male sex (coefficient 6.18, 95% CI 1.40–10.96; $p=0.01$), high social risk (coefficient –6.02, 95% CI –11.93 to –0.05; $p=0.046$), and neurodevelopmental diagnosis (coefficient –20.61, 95% CI –32.10 to –9.12; $p<0.01$); expressive communication (PLS-5) by high social risk (coefficient –9.30, 95% CI –13.89 to –4.71; $p<0.01$); and total language (PLS-5) by male sex (coefficient 4.54, 95% CI 0.25–8.83; $p=0.04$), high social risk (coefficient –8.15, 95% CI –13.36 to –2.94; $p<0.01$), and neurodevelopmental diagnosis (coefficient –16.42, 95% CI –27.53 to –5.31; $p=0.01$).

DISCUSSION

In our study, children born preterm demonstrated significantly higher rates of language difficulty but performed similarly to term-born children on voice and speech outcomes. Within the group born preterm, predictors of language, speech, and voice outcomes included high social risk, neurodevelopmental disorder, male sex, and chronic lung disease.

Our study provides an unprecedented depth of detail exploring language, speech, and voice outcomes at 3 years of age in children born preterm, in a large prospective cohort study with a comparison group of term-born children. We have reported data on the cooccurrence of speech, language, and voice difficulties, critical for clinicians and researchers to understand the overall communication profile of children born preterm.

The similarity between groups on voice outcomes may reflect improvements in respiratory care for neonates.¹⁰ Our finding that speech sound production does not differ between children born preterm and at term at 3 years contradicts previous findings,⁵ which may be attributable to our use of more detailed measures. Furthermore, similarities in voice and speech outcomes across children born preterm and at term also indicate that dysarthria (a disorder of the neuromuscular execution of speech that can affect articulation and voice) was not a significant issue in our population of children born preterm. This may be because children with known genetic neurodevelopmental disorders were excluded from the study at birth, and because the rates of significant brain injury and cerebral palsy were low in our cohort; also reflected in the low rates of oromotor difficulties (6% of the preterm group).¹⁴ The high rates of speech difficulties reported here (approximately 30% in the term-born group vs approximately 11% in other studies)⁷ suggest that the developmental norms provided by the DEAP did not reflect typical development in the population studied, and further work may be required to determine the reason for this.

Our use of a more psychometrically rigorous and detailed measure of language (PLS-5) than those used in other studies (e.g. Bayley Scales of Infant Toddler Development) lends additional credence to our findings with regard to low language difficulty rates and risk factors affecting language outcomes in this group. The language difficulties shown here, poorly predicted by medical factors, may support emerging arguments that the linguistic environment of the neonatal intensive care unit is a powerful contributor to later language performance.²³ However, further research will be required to determine the contribution of linguistic deprivation in the neonatal period versus other possible predictors such as parent–child interaction style and exposure to early learning environments.

Our findings indicate that children born preterm vary substantially in their communication profiles. It seems that some, but not necessarily all, children born preterm are at heightened risk of language, speech, and voice difficulties. Independent predictors of language, speech, and voice outcomes included a history of chronic lung disease (voice), male sex (language), neurodevelopmental disorder (language), and high social risk (language), many of which reflect established risk factors in the broader population.¹¹ Of these factors, high social risk is the most mutable. Therefore, one implication of our findings, is that more intensive surveillance and intervention may be best targeted to children born preterm at high social risk, in addition to those children with known sensory or neurodevelopmental diagnoses. Previous research has suggested that children born preterm are subject to a differential susceptibility effect – that is, they are more susceptible than term-

born children to the impact of their environment, positive or negative.²⁴ Thus, interventions to optimize the linguistic environment for high-risk children born preterm might be a particularly efficient allocation of resources.²³ This implication is especially pertinent given that our findings indicate that most children born preterm do not experience any language, speech, and voice difficulties at 3 years of age, thus directing more resources to the highest risk children may help to reduce unnecessary service provision. Future research may be directed at developing a model to estimate risk more precisely for this population, using a wider variety of risk factors such as days spent in hospital and home language environment.

A further risk factor that merits examination is oromotor feeding at 1 year. There has been an increasing interest in possible links between early oromotor performance and later speech and language outcomes.²⁵ In this study, oromotor feeding at 1 year of age was not found to be a statistically significant predictor of any 3-year language, speech, or voice outcome; however the odds ratios (ORs) and CIs for expressive language (OR –5.46, 95% CI –11.04 to 0.13) and total language (OR –5.71, 95% CI –11.89 to 0.48) suggest that this relationship warrants further examination.

One limitation of this study was the relatively low number of children/families who were able to complete all components of the assessment. This may reflect the need for different assessment tools in this population, to support fluctuating attention and engagement in standardized, ‘table-top’ assessment tasks, as well as the advisability of including assessments of broader neurodevelopmental functioning to inform factors that might contribute to participants’ assessment performance.

CONCLUSION

Children born before 30 weeks’ gestational age are at heightened risk of poorer language outcomes. Within the preterm group, children with a neurodevelopmental diagnosis, male children, and children at high social risk obtained lower language scores. Further investigation using similarly in-depth measures of language may inform a risk model that will assist in directing resources, such as language enrichment in the neonatal intensive care unit, to those children at greatest risk and who may respond to intervention most favourably.

ACKNOWLEDGEMENTS

We acknowledge the VIBeS team, which contributed to recruitment, data collection, and analysis; the families and children who participated in this study; and Diane Zannino, who provided statistical consultation. Funding for this study was received from the National

Health and Medical Research Council (AM Career Development Award #607315 and Practitioner Fellow #1105008; AS Career Development Fellowship 1108714, Centre for Research Excellence in Newborn Medicine grant 1060733; KS Centre for Research Excellence in Newborn Medicine grant 1060733), an Australian Government Research Training Program Scholarship (KS), and Speech Pathology Australia's Nadia Verrall Memorial Research Grant (KS). Murdoch Children's Research Institute research is supported by the Victorian government's Operational Infrastructure Support Program.

Supporting information

The following additional material may be found online:

Figure S1: CONSORT recruitment flowchart.

Table S1: Univariable analysis of predictors of communication outcomes in children born before 30 weeks

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Table 1: Participant characteristics

	Preterm (n=112)	Term (n=105)
Mean (SD) gestational age at birth (wk)	27.83 (1.44)	40.04 (1.19)
Mean (SD) birthweight z-score	-0.44 (1.04)	0.08 (0.78)
History of chronic lung disease	34 (30)	0 (0)
Male sex	55 (49)	52 (50)
Multiple birth	49 (44)	2 (2)
High social risk	39/101 (39)	23/95 (24)
Multilingual	13/106 (12)	12/101 (12)
Diagnosis of neurodevelopmental disorder	6/106 (6)	1/101 (1)
Oromotor feeding concerns at one year	26/68 (38)	13/100 (13)
Upper respiratory tract infection on the day of assessment	28/106 (26)	24/101 (24)
Mean (SD) corrected age (y) at assessment	3.21 (0.20)	3.20 (0.20)

Data are n (%) unless otherwise stated. SD, standard deviation.

Table 2: Speech, voice, and language outcomes

	Preterm	Term born	Magnitude regression coefficient (CI)	p
Speech ^a				
Percentage consonants correct standard score (n=194)	9.79 (2.09)	9.71 (2.11)	0.07 (−0.55 to 0.68)	0.83
Number of consonants produced (n=179)	21 (19–23)	22 (20–23)	−0.70 (−1.48 to 0.08)	0.08
Number of phonological processes (n=194)	2 (1–3)	2 (1–3)	−0.01 (−0.47 to 0.46)	0.98
Voice ^b				
GRBAS grade (n=204)	0 (0–1)	0 (0–1)	0.13 (−0.06 to 0.31)	0.17
Total voice handicap score (n=190)	11 (8–15)	10.50 (8–14)	1.63 (−0.57 to 3.84)	0.15
Language ^c				
Auditory comprehension (n=194)	97.23 (14.69)	101.82 (10.17)	−4.74 (−8.32 to −1.15)	0.01
Expressive communication (n=184)	98.54 (11.01)	105.27 (10.26)	−7.08 (−10.29 to −3.87)	<0.01
Total language (n=184)	98.00 (12.45)	103.95 (10.18)	−6.48 (−9.92 to −3.04)	<0.01

Data are mean (standard deviation) or median (interquartile range). ^aAssessed using the Diagnostic Evaluation of Articulation and Phonology. ^bAssessed using the Grade, Roughness, Breathiness, Asthenia, Strain voice rating scale and the Pediatric Voice Handicap Index. ^cAssessed using the Preschool Language Scales, Fifth Edition; and parent survey. CI, confidence interval.

Table 3: Multivariable analysis of predictors of communication outcomes in children born at less than 30 weeks' gestation

	Gestational age at birth	Male sex	Chronic lung disease	High social risk	Neurological diagnosis
Speech (DEAP)					
Number of consonants produced	0.38 (-0.02 to 0.78) p=0.06		-1.18 (-2.75 to 0.38) p=0.14		
Voice (pVHI)					
Total voice handicap score	-1.07 (-2.14 to -0.00) p=0.05		6.05 (1.21-10.89) p=0.02		
Language (PLS-5)					
Auditory comprehension		6.18 (1.40-10.96) p=0.01		-6.02 (-11.93 to -0.05) p=0.046	-20.61 (-32.10 to -9.11) p<0.01
Expressive communication				-9.30 (-13.89 to -4.71) p<0.01	-10.48 (-23.74 to 2.78) p=0.12
Total language		4.54 (0.25-8.83) p=0.04		-8.15 (-13.36 to -2.94) p<0.01	-16.42 (-27.53 to -5.31) p=0.01

All confidence intervals are 95%; shaded cells indicate that this relation was not significant on univariable analysis. DEAP, Diagnostic Evaluation of Articulation and Phonology; pVHI, Pediatric Voice Handicap Index; PLS-5, Preschool Language Scales, Fifth Edition.

Figure 1: Overlap between areas of communication impairment for children born preterm who completed all assessments (n=76). Thirty-seven children had no communication impairment.

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