Linguistic features in children born very preterm at preschool age

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ABBREVIATIONS

VPT Very preterm

AIM This cross-sectional study focused on the effect of very preterm (VPT) birth on language development by analysing phonological, lexical, grammatical, and pragmatic skills and assessing the role of cognitive and memory skills.

METHOD Sixty children (29 males, 31 females) born VPT (<32wks) aged 5 years were compared with 60 children with typical development. The linguistic assessment was performed by administering a battery of Italian tests for the evaluation of language; cognitive and memory skills were assessed by Raven's coloured progressive matrices and digit span subtest (Wechsler Intelligence Scale for Children [WISC-III]).

RESULTS Children born VPT showed delays in lexical (comprehension: *z*-score difference –1.18; 95% confidence interval [CI] –1.60 to –0.77; naming: –0.88; 95% CI –1.19 to –0.58) and pragmatic skills (comprehension: –0.76; 95% CI –1.02 to –0.49; narrative production: –0.47; 95% CI –0.72 to –0.23). Delays in phonology and grammar were less diffuse, involving productive skills (–1.09; 95% CI –1.64 to –0.54; –0.48; 95% CI –0.85 to –0.12, respectively), and were dependent by cognitive and memory skills. Lexical delays were more specific.

INTERPRETATION The linguistic profile of children born preterm is characterized by some abilities more impaired than others. This highlights the need of a linguistic assessment at the end of preschool age in order to plan a focused intervention aimed at improving lexical and pragmatic skills.

Recent reviews and meta-analyses suggest that very preterm birth (VPT, gestational age <32wks) might affect language development from the first years of life up to adolescence. 1-3 It has been argued that the developmental pathway of children born preterm is atypical and not merely delayed. Indeed, it is characterized by peculiar developmental patterns and relationships among competences consequent to the complex interaction between biological and environmental constraints linked to the premature interruption of pregnancy.2 However, to the best of our knowledge, the hypothesis of an atypical pathway in language development among preterm children has not been verified, and several questions still need answers. Is language development delayed or are some specific linguistic abilities impaired in preterm children? What are the roles of cognitive and memory abilities?

Previous studies have usually adopted global linguistic indexes,³ and have provided heterogeneous findings.¹ The few studies that have investigated specific linguistic abilities in children born VPT have usually focused on a restricted set of abilities. As a consequence, some linguistic skills, such as lexical processing,³ have been investigated far more

than others, such as phonological and pragmatic skills.^{1,2} The effects of preterm birth can also vary according to the child's developmental age.3 VPT infants have difficulties in a wide range of linguistic skills in the first years of life, with a higher incidence of language delay than in full-term peers: 34% at 3 years 6 months⁴ and 29% at 4 years of age. The few studies that have analyzed linguistic processing in 4- to 6-year-old children born VPT suggest that, by preschool age, difficulties become less diffuse and with contrasting results, which suggests that some linguistic abilities are more affected than others (Table SI, online supporting information). 6-10 However, no study has provided a comprehensive analysis of the effects of preterm birth on all linguistic skills. This is particularly true for pragmatic and discourse abilities: these skills have not been investigated among children born VPT at preschool age, whereas some difficulties have been described at 7 years and 9 years of age. 11,12

Another relatively unexplored issue relates to the role of cognitive and memory skills in language development. Studies on children with typical development have revealed that short-term memory, working memory, and fluid intelligence

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are separate constructs, even if highly related in young children, with different roles in supporting language acquisition (e.g. short-term memory linked to vocabulary, cognitive control to syntax). 13,14 The role of phonological short-term memory, working memory, and cognitive skills, as separate constructs supporting language development, has not been specifically investigated in children born VPT at the end of preschool age. This topic is particularly relevant in children born VPT who are frequently characterized by impairments in multiple domains.⁵ As revealed by the neuroconstructivism approach, 15 domains are not isolated from one another in their developmental trajectories, which suggests that one atypical ability can have ramifications on others. To our knowledge, some studies have investigated the role of cognitive ability on language development in children born VPT, and have shown contrasting results: in some studies, linguistic deficits persisted after controlling for cognitive scores; 8,9,16 in other studies, linguistic delays were explained by differences in general cognitive scores.¹⁷ Concerning working memory, a tight relationship with grammar has been described in children born VPT at 3 years 6 months. 18

To the best of our knowledge, this is the first study specifically designed to analyze the effects of very preterm birth on linguistic abilities at 5 years of age, taking into account potential differences in receptive and expressive abilities on phonological, lexical, grammatical, and pragmatic skills and analysing whether differences between children born VPT and children with typical development are related to their cognitive and memory difficulties. We hypothesized an atypical pathway in children born VPT, with some abilities more affected than others, and with different roles of phonological short-term memory, working memory, and other cognitive skills on language delay. We decided to investigate language development at the end of preschool age for the relevance of this developmental period. In fact, were we to reveal persisting difficulties at the end of preschool age, this would highlight how preterm birth not only causes difficulties in the first years, but also gives rise to a persistent delay. In addition, a basic knowledge of the effects of preterm birth on specific aspects of language processing at preschool age is particularly relevant when it comes to identifying children born preterm at risk of developing learning impairments at school age and to planning focused interventions.¹⁶

METHOD

Participants

One hundred and twenty monolingual Italian-speaking children were included in the present cross-sectional study. All participants were selected among middle class families of North Italy regions.

The first group comprised 60 children born VPT (<32wks gestational age). The comparison group comprised 60 children with typical development.

Children born preterm (29 males, 48%; 31 females, 52%) were born in 2007 and 2008 in the Neonatology Unit at Bologna University. Their mean age was 5 years 4 months (SD 4mo). Preterm children were recruited if, at

What this paper adds

- Very preterm children showed linguistic delays at 5 years of age.
- Impairments are evident in receptive/productive lexical and pragmatic skills.
- In phonology and grammar, only productive skills are compromised.
- Follow-up programmes should include a detailed evaluation of language.

birth, they had no major cerebral damage, no congenital malformations, and no indication of visual or hearing impairment. The characteristics of the preterm sample are presented in Table I. The children born preterm enrolled represented 83% of the eligible population (n=72; mean gestational age 28.9wks [SD 2.2; range 23-31]; mean birthweight 1195g [SD 376; range 509-1990]). No significant differences in gestational age or birthweight were found between children born VPT recruited (n=60) and not recruited (n=12; mean gestational age 29.2wks [SD 1.6; range 26-31]; mean birthweight 1222g [SD 425; range 626-1990]), because parents did not give their consent to enter into the follow-up programme.

All children with typical development (35 males, 58%; 25 females, 42%) had experienced normal birth and had no history of cerebral damage and/or congenital malformations or visual or hearing impairments. They were recruited from kindergartens in Northern Italy (mean of age at assessment 5y 4mo [SD 4mo]).

The two groups were comparable in terms of age at assessment, t(118)=-0.4; p=0.71, and sex, χ^2 (1, N=120) =1.2; p=0.27.

Procedure

The linguistic and cognitive assessment of the children born VPT was performed in a quiet room at the Day Hospital of the Neonatology Unit. Children with typical

Table I: Characteristics of the very preterm sample

	Very pre (n=60)	term group
Gestational age (wks), mean (SD) range	28.9	(2.29) 23–31
Birthweight (g), mean (SD) range	1177.53	(360.26) 509-1890
ELGA, n (%)	16	(27)
ELBW <1000g, n (%)	20	(33)
Caesarean section, n (%)	55	(92)
Multiple births, n (%)	22	(37)
SGA, n (%)	6	(10)
RDS, n (%)	52	(87)
Apnoea, n (%)	11	(18)
MV, n (%)	15	(25)
BPD, n (%)	13	(22)
IVH I/II grade, n (%)	2	(3)
ROP I/II, n (%)	13	(22)
Sepsis, n (%)	7	(12)
Mean length of hospitalization (d), mean (SD), range	55	(35.4) 6–152

ELGA, extremely-low gestational age (<28wks); ELBW, extremelylow birthweight (<1000g); SGA, newborn infants with birthweights <10th centile for gestational age, according to North Italian growth charts (http://www.inescharts.com/); RDS, respiratory distress syndrome; MV, mechanical ventilation; BPD, bronchopulmonary dysplasia; IVH, intra-ventricular haemorrhage detected by cerebral ultrasound scan; ROP I/II, retinopathy of prematurity of grade I or II.

development were assessed in a quiet room at their school. All children were assessed by trained psychologists.

The study protocol met the ethical guidelines for protection of human participants, including adherence to the legal requirements of the country, and received a formal approval by the local ethics committee (Comitato Etico Indipendente dell'Azienda Ospedaliero - Universitaria di Bologna, Policlinico S. Orsola-Malpighi). The parents of children released their informed written consent for participation in the study, data analysis, and for anonymous data publication.

Materials

The linguistic assessment was performed by administering a selection of tests from the 'Batteria per la Valutazione del Linguaggio in Bambini dai 4 ai 12 anni' (BVL 4-12).¹⁹ This is a comprehensive battery of tests with solid theoretical and psychometric properties that has been designed to assess comprehension, oral production, and repetition skills in children aged 4 through to 12.19, 20 For the direct purposes of the current study, we focused on tasks assessing phonetic and phonological processing, lexical, grammatical, and pragmatic skills.

Phonetic and phonological skills

Phonological discrimination: The children listened to pairs of identical items or minimal pairs. Percentage of accuracy was calculated.

Articulatory skills: The children were administered a naming subtest (see 'Naming', below). Two points were assigned for each word that had been correctly produced at the first presentation and one point for each word that had been correctly repeated after the examiner presentation (maximum score=154).

Lexical skills

Lexical comprehension: The children were asked to identify which, among four pictures, represented the meaning of a word uttered. One point was assigned for each correct answer (maximum score=18).

Naming: The children had to name a series of stimuli depicted in sheets. For each correct answer, one point was assigned (maximum score=77).

Grammatical skills

Syntactic comprehension: The children were asked to identify which, among four pictures, represented the meaning of a sentence uttered. One point was assigned for each correct answer (maximum score=40).

Sentence completion: The children listened to a sentence, then heard the beginning of a second sentence, and were required to assign the correct morphemes to the verb in the second sentence. One point was assigned for each correct answer (maximum score=14).

Pragmatic skills

Comprehension of idiomatic expressions: The children listened to idiomatic sentences and decided which of three potential meanings was correct. Each correct answer was assigned one point (maximum score=10).

Global coherence in narrative production task: The children described a cartoon story made of six drawings, appearing on the same page. The examiner explained that (s)he was not familiar with its content. Errors of global coherence were analysed, including the production of utterances that may be tangential, conceptually incongruent with the story, propositional repetitions, or simple fillers. The percentage of utterances without errors of global coherence was computed.

Cognitive skills

Raven's coloured progressive matrices (CMP)²¹ as a measure of non-verbal cognitive development.

Digit span subtest (forward and backward) of the Wechsler Intelligence Scale for Children (WISC-III)²² as a measure of working memory.

Non-word repetition task of the BVL 4-12^{19,20} as a measure of phonological short-term memory. Each correct repetition received one point (maximum score=15).

Statistical analyses

Statistical analyses were run using SPPS 21 (IBM Corp., New York, NY, USA). The significance level was set at 0.05. Cognitive and linguistic scores are reported as means and standard deviations or proportions. Analyses of variance (ANOVAs) were run to investigate differences between VPT and children with typical development in all cognitive and linguistic measures. Analyses of covariance (ANCOVAs) were run in order to compare linguistic abilities between VPT and children with typical development, adding as covariates the scores obtained by tests of cognitive non-verbal development, digit span, and non-word repetition.

RESULTS

Cognitive (Table II) and linguistic (Table III) scores are reported as means and SDs or proportions, where appropriate. The children born preterm had significantly lower scores than their peers in Raven's matrices, digit span, and non-word repetition (Table II).

In regards to language, preterm children had significantly lower scores in articulatory skills, lexical comprehension and naming, sentence completion, comprehension of idiomatic expressions, and global coherence in the narrative production task (Table III). No significant grouprelated differences were found in phonological discrimination and syntactic comprehension. Figure 1 shows the standardized differences (mean of z-scores) between children born VPT and children with typical development. For each child born VPT, the z-score was given by the child's score minus the mean score of the children with typical development, divided by the children with typical development's SD. Children born VPT had lower scores in all linguistic abilities than the comparison group (Fig. 1). Children born VPT scored less than −1SD in lexical comprehension (z=-1.18; 95% CI -1.60 to -0.77)

Table II: Summary of means and standard deviations (SDs) for cognitive abilities in the very preterm and comparison groups. F_r , p_r , and partial eta-squared (η_0^2) values for each ANOVA are reported

		Preterm grou	р	Co	mparison gr	oup		ANOVA	
	n	M	SD	n	M	SD	F	р	η_p^2
Non-verbal cognitive skills Raven's coloured progressive matrices ^a	60	15.2	3.4	60	19.3	5.0	27.7 ^b	<0.01	0.190
Memory Digit span ^a Non-word repetition ^a	60 60	5.9 11.2	2.0 3.8	60 60	9.0 13.3	2.6 2.1	24.6 ^b 14.2 ^b	<0.01 <0.01	0.172 0.107

Significant results are in bold. ^aRaw score (maximum scores where available are reported in the Method section). ^bWelch's *F* applied. M, mean.

and articulatory skills (z=-1.09, 95% CI -1.64 to -0.54), between -0.50 and -1SD in naming (z=-0.88; 95% CI -1.19 to -0.58) and comprehension of idiomatic expressions (z=-0.76; 95% CI -1.02 to -0.49) and between 0 and -0.5 in sentence completion (z=-0.48; 95% CI -0.85 to -0.12), global coherence in narrative production (z=-0.47; 95% CI -0.72 to -0.23), phonological discrimination (z=-0.35; 95% CI -0.63 to -0.07), and syntactic comprehension (z=-0.27; 95% CI -0.52 to -0.02).

Considering the relevant differences between groups in cognitive skills, we investigated whether the differences persisted after controlling for scores on Raven's matrices, digit span, and non-word repetition (Table III). As for articulation, the difference between VPT and children with typical development was still significant when Raven's score and digit span were added as covariates, although it disappeared after controlling for non-word repetition. The lower scores of children born VPT in lexical comprehension and naming remained significant after controlling for cognitive and memory scores. On the contrary, the grouprelated differences in sentence completion did not survive after covariation. As for pragmatics, group effect persisted after controlling for the covariates in the comprehension of idiomatic expressions, while in global coherence the difference between groups was significant when digit span and non-word repetition were added as covariates but disappeared after controlling for Raven's score.

DISCUSSION

Very preterm birth alters language development as well as non-verbal cognitive skills, working memory, and phonological short-term memory during preschool age. Its effects on language development are far from homogeneous. The detailed linguistic analysis revealed a profile with peaks and valleys: lexical and pragmatic abilities were particularly weak in both productive and receptive domains, whereas phonological and grammatical delays were less diffuse, affecting productive but not receptive skills. Some of these linguistic drawbacks were affected by the level of non-verbal cognitive abilities or memory, whereas others remained when controlling for these abilities. In addition, as revealed by Figure 1, language delays for children born VPT were not severe, because mean group differences with children

with typical development were comprised within -1SD. In our opinion, these findings have important theoretical and clinical implications and provide some new insights in the recent debate about the construct of language impairment in general²³ and the characteristics of linguistic development in children born VPT in particular. This is a relevant issue as a consensus on diagnostic criteria and terminology for unexplained language problems in children is still lacking.²³

The group of children born VPT had impairments in non-verbal cognitive skills, working memory, and phonological short-term memory. These findings are coherent with previous observations on the effects of preterm birth on cognitive development²⁴ and suggest that difficulties in working memory and phonological short-term memory, already detectable at 3 to 4 years of age, ¹⁸ are still observable at 5 years and may indirectly affect linguistic development. Notably, this study showed the presence of difficulties in a test of non-word repetition in children born VPT, which is considered a reliable marker of verbal difficulties in children with language impairments.²⁵

Preterm birth impacts language development at different levels, with some differences between receptive and productive skills. Severe difficulties were found in lexical comprehension and naming, which confirms previous findings that highlighted the presence of reduced receptive³ and productive lexical skills at preschool age. 5,8,9,17 Notably, the group-related difference in naming and lexical comprehension persisted even when non-verbal cognitive and memory skills were included in the models, although studies on children with typical development have shown that lexical development is strongly related to phonological short-term memory. 14 Overall, these findings suggest that the observed lexical difficulties reflect an atypical organization of the lexical system in children born VPT. The possibility of such an atypical organization has received some evidence from recent behavioural and experimental studies. Two-month-old infants born preterm showed a delay in sensitivity to synchronous syllable-object pairings. The authors hypothesized this result as an important index for delays in word mapping and comprehension because in natural naming contexts, caregivers use a great deal of synchrony between spoken words and object motions.²⁶ In a

Table III: Summary of means and standard deviations (SDs) for linguistic development in very preterm and comparison groups. F, p, and partial eta-squared (11,2) values for ANOVAs and ANCOVAs are reported

	>	Very preterm group	r.r.	ŭ	comparison group	u		ANOVA						ANCOVA	_			
	u	Σ	SD	u	Σ	SD	F	р	η_p^2	ਮੁ	$\rho^{\rm c}$	η_p^{2c}	쩐	$p^{\rm q}$	$\eta_p^2 d$	Fe	$p^{\rm e}$	η_p^2 e
Phonology																		
Phonological discrimination ^a	09	70.1	38.7	09	82.6	35.7	3.9	0.07	0.028	0.3	0.57	0.003	0.2	99.0	0.002	0.01	0.94	<0.001
Articulatory skills ^b	9	130.9	16.9	09	139.6	8.0	13.0 ^f	<0.01	0.100	9.9	0.01	0.053	2.7	0.02	0.047	3.2	0.08	0.027
Lexicon																		
Lexical comprehension ^b	9	13.4	3.0	09	15.6	1.9	23.2	<0.01	0.164	11.9	<0.01	0.093	12.1	<0.01	0.094	14.8	<0.01	0.113
Naming ^b	9	60.5	5.9	09	64.8	4.9	19.8	<0.01	0.143	11.5	<0.01	0.090	9.3	<0.01	0.074	13.3	<0.01	0.102
Grammar																		
Syntactic comprehension ^b	09	24.6	8.3	09	26.9	9.8	2.3	0.14	0.019	0.01	0.90	<0.001	0.01	0.90	<0.000	0.03	98.0	<0.001
Sentence completion ^b	09	7.0	3.0	09	8.0	2.1	4.4₊	0.04	0.036	1.1	0.30	0.009	9.0	0.43	0.002	8.0	0.37	0.007
Pragmatics																		
Comprehension of idiomatic expressions ^b 60	09	1.6	1.6	09	2.7	1.5	16.8	<0.01	0.124	12.3	<0.01	0.095	6.6	<0.01	0.078	10.5	<0.01	0.082
Global coherence ^a	09	77.1	14.7	09	84.4	15.5	7.2	0.01	0.057	2.4	0.12	0.020	4.2	0.04	0.035	8.9	0.01	0.055

Significant results are in bold. *Per cent accuracy score; baw score (maximum scores are reported in the Method section); Covariate: non-verbal cognitive skills, Covariate: digit span, Covariate: non-word repetition. Welch's Fapplied. M, mean.

recent functional magnetic resonance imaging study, Schafer et al.²⁷ analysed the neural activations elicited in a group of adolescents born VPT during a visual semantic association task. From a behavioural point of view, no group-related differences were found. However, children born VPT had a significant decrease in left frontal and bilateral temporal white matter volumes and engaged different neural pathways. In comparison participants, the semantic association task activated bilateral inferior frontal and middle temporal gyri; in children born preterm, the semantic association recruited the functional association of temporal areas with activation in the supplementary motor area. Further research should investigate the neural underpinnings of semantic processing in preschool-age children born VPT and the role potentially played by their motoric difficulties²⁸ in language development. As recently suggested by Sansavini et al.²⁹ difficulties in motor development in the first years of life may affect the construction of meaning and the organization of semantic representations, delaying their lexical development.

This central difficulty in semantic representation might also indirectly affect other skills, such as pragmatic abilities. Our study highlighted the presence of difficulties in the ability to understand non-literal expressions and to generate coherent discourse. Previous studies suggest that pragmatic and discursive abilities are impaired in these children at school age. 11,12 To the best of our knowledge, this is the first evidence of pragmatic difficulties at preschool age.

Concerning the comprehension of idiomatic expressions, the group-related difference persisted after controlling for measures of non-verbal cognitive and memory functioning. This suggests that, at least at this age, idiomatic expressions might be stored as complex linguistic representations with idiosyncratic meanings in the lexicon.³⁰ Therefore, together with the already observed lexical difficulties, the reduced comprehension of idiomatic expressions might further confirm a specific difficulty in the ability to select appropriate lexical items in the mental lexicon and to construct complex meanings. Another consideration relates to the difficulty encountered by children born preterm in producing samples of narrative language with adequate levels of global coherence. The group-related difficulties in maintaining adequate levels of global coherence accuracy were no longer significant when their level of non-verbal cognitive functioning was taken into account. This finding was expected. Indeed, several studies suggest that the ability to monitor, inhibit, and control the flow of thoughts that characterize the process of generation and production of narrative discourse depends on high-level cognitive skills.31

Previous investigations showed that children born preterm have difficulties in phonological discrimination at 4 years.6 Our study suggests that this difficulty is no longer observable at age 5 because these children apparently learn how to cope with this critical aspect of language processing. However, they continue to experience articulatory difficulties. This was expected, because previous studies

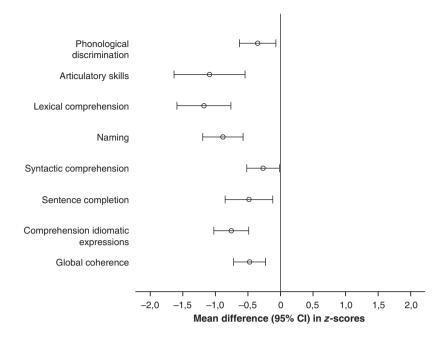


Figure 1: Mean z-scores and 95% CIs for children born VPT in relation to the comparison group on linguistic skills.

showed delayed articulatory development in children born VPT¹⁰ and even more in those born extremely preterm¹⁷ at 6 years of age. This further supports the idea of a complex interaction between motoric and linguistic development in these individuals. Interestingly, the group-related difference in articulatory skills was no longer significant when the score at the non-word repetition task was added as a covariate. Because this task involves not only phonological short-term memory skills but also perceptual and motoric abilities,³² the role of motor skills seems particularly relevant in language development in children born preterm.

Contrary to previous findings,^{7,9} the children born VPT included in this study did not experience difficulties in syntactic comprehension. Furthermore, although they had difficulties in grammar productive skills, 10 the group-related difference in the sentence completion task was no longer significant when non-verbal cognitive and memory skills were added as covariates. This confirms the important role played by these cognitive functions in the development of grammar, similar to previous findings of studies on children with typical development¹⁴ and on children born VPT.¹⁸ Further cross-sectional and longitudinal studies are necessary to explicitly assess the relationship between cognitive and grammatical skills in these children, and between lexicon and grammatical skills. Previous studies have revealed tight relationships between lexicon and grammar in children born VPT,9 while in the present study lexicon and grammar development appear to be affected differently by preterm birth.

Some limitations of the current study need to be taken into account, as both suggestions for future studies and cautions in the interpretation and generalization of our results. First, measures of attention and executive functions would be useful to better understand the linguistic profile of children born preterm, because difficulties in selective and sustained attention and executive functions are not unusual among preterm children.³³ Further studies should explicitly address this issue. Second, in the present study, standardized tests were employed to describe the linguistic profile of children born preterm. Future studies should associate standardized tests with experimental tasks and neuroimaging measures in order to understand the processes underlying the delayed skills. Third, this cross-sectional study described a detailed picture of linguistic development in preterm children at just one point of observation, at the end of preschool age. Further longitudinal studies are needed to describe the developmental trajectories of language development in very preterm children from the first years of life. Such studies will provide relevant information about the relationships among the different linguistic skills, allowing for an early detection of potential risks in language development that, if unrecognized, might affect the development of higher-level linguistic skills. These efforts will certainly trigger the ideation of early intervention protocols that might take place even earlier than preschool age.

In conclusion, the results from this study allow us to draw two major conclusions. From a theoretical point of view, they show that the profile of preterm children is not characterized by a general delay but by an atypical developmental trajectory with some degree of intra-group variability: some linguistic abilities are more affected than others, and are apparently linked to delays in other domains such as cognition and verbal working memory. According to the neuroconstructivist approach, the present study shows a continuity between pre- and perinatal life and subsequent

development, suggesting that the biological and socioenvironmental constraints associated with preterm birth, which occurs in a critical period of rapid development of the neural system, lead to an atypical development.² From a clinical point of view, the current study demonstrates the need for a detailed linguistic assessment in children born preterm in order to detect those at risk for future linguistic impairments and plan effective intervention programmes focusing on specific abilities such as lexical and pragmatic skills. A 'wait and see' strategy is dangerous and inappropriate for children born preterm: a detailed evaluation and early intervention at preschool age may decrease the incidence of delays when children reach school age and thus support preterm children in the transition to literacy.

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SUPPORTING INFORMATION

The following additional material may be found online:

Table SI: Summary of articles including the analysis of specific language subdomains, ordered by the age of evaluation from 4 to 6 years of age.

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