



Validity of the language development survey in infants born preterm



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ABSTRACT

Background: Preterm infants are at greater risk of language delay. Early identification of language delay is essential to improve functional outcome in these children.

Aim: To examine the concurrent validity of Rescorla's Language Development Survey and the Bayley Scales of Infant and Toddler Development (Bayley-III) at 18 months corrected age in preterm infants.

Study design: Test accuracy study.

Participants: 189 preterm infants born <29 weeks were assessed at 18 months.

Outcome measures: The Language Development Survey, a parent-reported screening instrument, was administered in French concurrently with the Language Scales of the Bayley-III. Receiver-Operating-Characteristics curves were used to determine optimal cut-off score on the Language Development Survey to identify Bayley-III score <85. Sensitivity, specificity, positive and negative predictive values, and κ coefficient were calculated.

Results: Using Rescorla's original cut-off scores of ≤ 10 words for boys and ≤ 24 for girls, sensitivity was 76% and 88% for boys and girls, respectively, and specificity was 73% and 52% for boys and girls, respectively, in identifying language delay as per the Bayley-III. The optimal threshold was ≤ 10 words for both boys and girls. In girls, lowering the cut-off score decreased sensitivity (79%), but improved specificity (82%), thus lowering the number of false-positives.

Conclusion: Our findings support using the Language Development Survey as an expressive language screener in preterm infants.

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1. Introduction

Communication and language problems are frequently encountered in infants born prematurely [1]. More specifically, preterm children exhibit difficulties across all language sub-domains, including phonological awareness, semantics, grammar, discourse, and pragmatics [2,3]. Language difficulties persist throughout the school years with 15% of preterm children still requiring speech therapy at 12 years [4]. The first cues indicating language problems can be identified as early as during infancy. Indeed, among extremely preterm infants, 20–35% have language delay according to the Bayley Scales of Infant and Toddler Development 3rd edition (Bayley-III) at 18–24 months corrected age (CA) [5].

Language impairment has a significant long-term impact and may affect social relationships, literacy and educational attainment [6]. Therefore, early recognition of language delay is crucial to improve functional

and academic outcomes of at-risk children. As such, the American Academy of Pediatrics has been advocating for standardized developmental screening at specific ages using validated tools including parent-reported questionnaires [7]. Prompt identification of language developmental delay allows the child to benefit from early speech and language intervention which has been shown to enhance language abilities and to reduce the burden of a persistent disability on child's function [8].

Standardized language-specific screening tools for infants that are parent-administered and quick to complete and score are scarce. One such parental questionnaire, the MacArthur-Bates Communicative Development Inventory (CDI), has been adapted in multiple languages and widely used for research with preterm populations in several countries [9–14]. At 12–24 months corrected ages, preterm infants display lower gesture/word production, as measured by the CDI, as well as smaller vocabulary size and shorter length of utterances compared to term controls, with lower gestational ages associated with more impairment [9,15,16].

Though the CDI has proven useful to assess language development in preterm infants, time required to complete the form (30–40 min) and to score it (20 min) is rather long and could hinder uptake by busy general health care providers. To overcome these barriers, short-form versions of the CDI were created that consist of an 89-word checklist for

Abbreviations: LDS, Language Development Survey; CA, corrected age.

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vocabulary comprehension and production at 8–18 months and of a 100-word checklist for vocabulary production at 16–30 months and a question on word combinations [17]. These have been adapted in several languages [18–21], but not validated in preterm populations yet. The Language Development Survey (LDS), another vocabulary production checklist filled by parents, is also a shorter alternative (<10 min to complete and <5 min to score) that may be easy to use in practice [22]. The LDS has been recommended as a valuable tool to screen specifically for language delay in the general population [23]. In addition, it has been validated in bilingual homes in the United States using the English version of the questionnaire [24]. However, data in preterm infants remain limited. Only Mossabeh et al. compared results obtained from the LDS with the Bayley-III language composite score in preterm infants aged 22 to 26 months and showed a sensitivity of 61% and a specificity of 82% [25]. Psychometric properties of the LDS have not yet been determined at an earlier age in preterm infants.

Therefore, this study aimed to examine the concurrent validity of the LDS compared to the language composite score of the Bayley-III in preterm infants born <29 weeks of gestation at 18 months CA. This cohort consisted of French-speaking families. The 18-month visit is a key encounter to conduct developmental screening as recommended both by the American Academy of Pediatrics and the Canadian Pediatric Society [7,26]. We postulated that the LDS would be a reliable screening tool that is able to discriminate with an 80% sensitivity and specificity preterm born infants with language developmental delay.

2. Material and methods

2.1. Participants

This was a retrospective cross-sectional analysis of a cohort of infants prospectively followed from neonatal discharge until 18 months corrected age. All infant survivors born <29 weeks of gestation and admitted to Centre Hospitalier Universitaire Sainte-Justine neonatal intensive care unit were eligible to be seen at the neonatal follow-up clinic as part of routine developmental surveillance and screening. There were no exclusion criteria for follow-up. For the purpose of this current study, we included all infants born between April 2009 and December 2012, aged 17 to 19 months corrected age at time of follow-up visit, who were able to complete the Bayley Scales of Infant and Toddler Development third edition (Bayley-III) and whose parents had filled the Language Development Survey (LDS). The local institutional ethics board approved of the study.

2.2. Procedure

As part of the 18-month CA visit routinely offered to preterm infants born <29 weeks of gestation, all children were administered the Bayley-III which was performed in French by a trained psychologist who was blind to prematurity-related complications and results of the LDS (see below for details on the Bayley-III and LDS). Indeed, the psychologist, who was meeting with the family for the first time, only reviewed medical charts after completion of testing. Afterwards, regardless of performance on developmental testing, all parents were asked to fill the French version of the LDS. If the child said words in another language, parents were told to report them as well as described by Rescorla et al. [24]. Although they were present during Bayley-III administration, they were unaware of Bayley-III scores while completing the LDS. Birth history and perinatal data were obtained from the infant's medical record. Parents were also asked about their educational level, living arrangements, and child language exposure. Finally, a pediatrician completed the physical and neurological examination. Due to the retrospective nature of the study, we could not record reasons why children could not be tested on the Bayley-III or why parents did not fill the LDS.

2.3. Index test

Rescorla's LDS is a 310-word checklist used to identify language delay in 18–35 month-old children [22]. Parents are asked to mark all words spontaneously said by the child and to describe word combinations. Additional questions inquire on number of otitis media, number of languages spoken at home, family history of language delay, and parental concerns regarding child's language development. The total number of single words represents the infant's score. Above 24 months, length of word combinations is also taken into account. For infants between 18 and 23 months, a score ≤ 10 words for boys and ≤ 24 words for girls (≤ 15 th percentile) suggests expressive language delay. Strong reliability has been demonstrated with test-retest reliability and Cronbach's alpha internal consistency of both 0.99 [22,24]. Correlation between 0.72 and 0.87 with other vocabulary tests indicates good validity [24,27]. Sensitivity is $\geq 80\%$ and specificity $\geq 85\%$ [24]. Completion takes <10 min and requires a fifth-grade level reading skill.

2.4. Reference test

The Bayley-III is a widely used and validated measure of child's developmental status which yields three main scales: cognitive, language and motor [28]. The language composite score combines results obtained from the expressive and receptive scaled scales. Mean is set at 100 with a standard deviation of 15. Language delay is defined as a language composite score < 85. Reliability is above 0.80. Validity measures range between 71% and 83% for the language scale when compared to the Wechsler Preschool and Primary Scale of Intelligence third edition (WPPSI-III) [28].

2.5. Statistical analysis

Descriptive statistics were computed in terms of means, standard deviations, and frequencies. Receiver-operating-characteristic (ROC) curves were constructed for boys and girls separately to find the optimal cut-off value on the LDS to identify a Bayley-III language composite score < 85. We calculated Cohen's kappa to determine chance independent agreement between classifications (i.e. normal language vs. language delay). Concurrent validity of the LDS in identifying children with Bayley-III language composite score < 85 was further assessed with sensitivity, specificity, positive predictive value, negative predictive value and 95% confidence interval. Statistical analyses were performed using SPSS 21.0.

3. Results

3.1. Descriptive analyses

Of 350 survivors born <29 weeks gestation, 289 (83%) attended their routine follow-up visit at 18 months CA, but 18 were outside the age range (>19 months CA) and therefore excluded leaving 271 eligible infants (Fig. 1). Complete dataset for both the Bayley-III and the French version of the Language Development Survey (LDS) was available for 189 of them. Participants did not differ from non-participants in terms of gestational age, birth weight, sex, oxygen use at 36 weeks CA, severe retinopathy of prematurity, severe brain lesions, duration of neonatal hospitalization, household arrangement, maternal age and maternal ethnicity. However, participants were more likely to be singletons (81% vs. 55%) with mothers with an educational level above high school (67% vs. 50%).

Developmental assessments were conducted at a mean corrected age (CA) of 18.2 months (range: 17 to 19 months). Sixty-nine percent of mothers described themselves as Caucasians. For the majority of families, primary language spoken at home was French (89%) with one third of children exposed to a bilingual environment. A high proportion of parents (43%) expressed concerns regarding their child's language

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