INTRODUCTION

In Sweden, language disorder is identified by evidence-based language screenings within the Child Health Services (CHS) framework. In some regions, this takes place when children are 2.5 years old and in other regions at 3 years. After referral to a speech and language pathologist (SLP), 87% of the positively screened children are diagnosed with developmental language disorder. For children below age 2.5/3 years, questions to parents on communication and language milestones are part of the developmental surveillance programme.
Each occasion is pre-planned and regulated, enhancing the surveillance precision compared with relying on the judgement of individual professionals’ or parents’ concerns. Unfortunately, the validity of this language surveillance has not been investigated. Attempts to evaluate formal screening procedures below 2.5/3 years have been conducted, but satisfying psychometric properties have not been reported. A recent publication by the Swedish National Board of Welfare concluded that research about early screening for language and communication delay is still insufficient. In an international perspective, there is no consensus on the use of general language screening for children in preschool years.

Babbling develops through several stages during the first year of life. The infraphonological model highlights how early vocalisations gradually develop to be more speech-like and thereby prepare the child for speech. During the canonical babbling (CB) stage, children combine earlier practiced sounds into well-formed syllables, defined as a consonant and vowel with rapid transition in between, leading to the first word production. The CB onset occurs between 6 and 8 months and by the age of 10 months at least 93% of all children use CB. Established CB is considered a positive predictor of expressive speech and language development. In clinical groups, such as children with severe hearing loss, unrepaired cleft palate or neurodevelopmental disabilities, deviant CB has been described as a risk factor for later difficulties with expressive language, that is, with expressive vocabulary and consonant sound system proficiency. Despite this associated risk, CB is seldom included in screening studies. As a target for early detection CB has only been investigated in selected populations and, to our best knowledge, not in a community setting. In a high-risk population, Oller et al. performed a screening targeting CB, using validated open-ended parent questions. The screening outcome was confirmed in a laboratory and at follow-up at 30 months of age smaller expressive vocabulary was noted for children with late CB onset. The authors’ conclusion that CB might ‘provide an important new element in an effective screening battery’ (p.240) was the starting point of the current study where the validated open-ended questions to parents about their child’s babbling were used at the 10-month visit to CHS. The intended use of the babbling questions was as a valid surveillance of early language development, complementing current measures. The study objective was to evaluate the concurrent and predictive validity of the babbling questions in a non-selected population within the CHS. The first hypothesis was that the babbling questions at 10 months would identify most children without CB, reflecting a high concurrent validity. The second hypothesis was that the babbling questions could predict the outcome on the language screening 2 years later, reflecting a high predictive validity.

3 | MATERIALS AND METHODS

This prospective study consisted of three parts, targeting the same children at different ages. The babbling questions were asked when the children were 10 months, the babbling assessment was performed before 12 months of age and the routine language screening was performed when the children were 2.5 or 3 years of age. In this study, we have followed the guidelines for Standards for the Reporting of Diagnostic Accuracy Studies (STARD) and uploaded the STARD checklist as Supporting information S1.

The open-ended babbling questions, posed by the CHS nurse at the routine 10-month-visit, constituted the index test and consisted of one to three separate questions to parents. The questions were formulated to elicit parents’ own examples of their child’s babbling and there presented in Appendix 1.

The two pre-specified result categories were established CB and not CB and reflected if the child had reached the CB stage or not. The categories were based on the CHS nurse judgement of the babbling examples given by the parents and not on earlier knowledge of the child’s developmental history or clinical status. The questions were posed in hierarchical order, and the CHS nurse continued to the next question only if there were no examples of CB on the previous question. When examples of CB were given on any of the three questions, the notation established CB was made, and the child was considered to have reached the CB stage. Children with the notation not CB on all three questions were considered to not have reached the CB stage and were scheduled for the babbling assessment.

Children in the non-CB group took part in the babbling assessment conducted by a project SLP to confirm the lack of CB. On average the babbling assessment took place 44 days (SD = 25) after the babbling questions and consisted of approximately 45 min free play interaction between the child and a parent using age-appropriate toys. The parent instruction was to interact as usual. The session took place in a CHS centre or SLP office and was audio-video recorded with a camcorder HC-V750 (Panasonic Holding Corp), external microphone ECM-M5957 (Sony Corp) and for back-up, an external audio recorder DR-22WL (Tascam). The assessment goal was 100 child utterances, defined by breath groups and for children with fewer utterances, all recorded utterances were included. A well-trained SLP counted the utterances from the recordings and calculated a CB ratio by dividing the number of CB utterances by the total number of utterances. A CB ratio of 0.15 or more was considered to indicate established CB. The same SLP repeated the
counting and calculation on 27% of the children and another SLP on 40%, reaching 100% intra-rater agreement and 83% inter-rater agreement.

The language screening was chosen as the reference standard for calculation of the predictive validity as it is the method used for detection of children with language disorder within the CHS framework. The results of the language screening in the relevant regions, at 2.5 or 3 years, were collected by the child’s CHS nurse who had access to clinical information as well as the index result. The 2.5-year screening, relevant for 512 participating children, is based on structured play with toys and includes instructions, imitation, naming and use of two-word sentences.\(^1\) The 3-year screening, relevant for 614 participating children, is based on pictures of everyday objects and includes instructions, use of three-word sentences and level of co-operation.\(^2\) Both screening procedures aim to identify children with language delay by comparing their language abilities with age-relevant norms. According to the screening manuals, a passed screening was considered as typical language development and a failed screening as suspected language disorder, that is the target condition, leading to referral to an SLP for assessment and diagnosis.

Information about the research project was given to CHS centres in the Stockholm and Gothenburg regions, in Sweden, via CHS newsletters, large-scale lectures and repeated information meetings. There were 25 CHS centres that volunteered to participate, equally distributed in the two city regions. The CHS nurses recruited participants at the regular 8-month visit, children with at least one native Swedish-speaking parent were eligible for inclusion and were recruited consecutively. The recruitment period lasted from January 2015 to April 2016. The CHS centres differed in number of enrolled children (\(M=41.7\) children, range = 5–136). Figure 1 contains a flow-chart of all participants in the project. The parents of 1219 children consented to participate in the overall project, corresponding to 55% of those asked to participate. The parents of 93 children participated only by sharing the language screening result.\(^25\) The number of participating 10-month-olds (\(n=1126\)) roughly corresponds to 1/5 of the eligible population in the two city regions. Ethical approval was obtained from the Regional Ethical Committee in Stockholm (Dnr 2015/1401-31) and both parents provided informed written consent for their child to take part in the study.

### 3.1 Analysis

To evaluate if the babbling questions to parents identified children without established CB at the time of asking, the results for the non-CB group were compared with the babbling assessment results. The results were summarised as percent agreement. To evaluate if the babbling questions at 10 months could predict language development 2 years later, the results of the babbling questions were compared with the language screening results. These results were summarised with relevant predictive validity statistics. The screening results at 2.5 and 3 years were analysed separately, but as they were highly similar, the results are reported collapsed. There was no result that was hard to interpret, children with missing data on either index test or reference standard were excluded from the analysis of predictive validity.
4 | RESULTS

Of the 1126 children who took part, 16 (1.5%) had not established CB according to the babbling questions to parents at 10 months, a proportion of 1.4%. The mean age of the 16 children (nine boys) in the non-CB group was 10.4 months (SD 0.9). All the children in the non-CB group had passed the new-born hearing screening.26 four children had a birth complication and one child suffered from a medical diagnosis. Both parents of 11 children and one parent of three children had education above high school, and for one child information was lacking.

After one child dropped out, 12 of 15 (80%) children in the non-CB group were confirmed as lacking CB, indicating that the concurrent validity of the CB questions was high. There were no reported difficulties in administering the babbling questions, while four children did not cooperate fully in the language screening.

Of the 916 children who underwent language screening 2 years later, 71 (7.8%) failed the screening. Information on clinical interventions between the index test and the reference standard was not gathered. Among the 93 children who only shared the language screening result, eight (9%) failed the language screening.23

Table 1 includes a comparison of the babbling questions and the language screening, and Table 2 contains summary statistics regarding the predictive value of the babbling questions calculated from the frequencies in Table 1. In summary, in the non-CB group, the risk of a failing language screening 2 years later was higher than in the CB group (a relative risk of 5.54 and a positive predictive value [PPV] of 40%). Further, the babbling questions correctly predicted most children with later typical language development (specificity 99%) and the probability of having typical language in the group with established CB was high (negative predictive value [NPV] 93%). However, only six of the 71 children who failed the language screening were identified as lacking CB by the babbling questions (sensitivity 8%). The lack of CB was confirmed by the babbling assessment for all these six children. As an overall measure of predictive validity, the diagnostic odds ratio was 8.57.

5 | DISCUSSION

The babbling assessment confirmed a lack of CB for most children without CB according to the babbling questions, demonstrating a high concurrent validity of the babbling questions in the non-CB group. Three children in the non-CB group were assessed to have CB. Presumably, this was related to maturation during the time interval between the babbling questions and the babbling assessment. The result expands on results from earlier studies11,27 by showing that not only a specialist but also a generalist, such as a CHS nurse, can make a valid decision on CB after open-ended questions to parents.

The babbling questions are valid measures of babbling development. Accordingly, it is suggested that they can be included in the CHS language surveillance at 10 months to assess the child’s current babbling status. Today, questions regarding early vocalisation are asked at about 6 months and concerns so-called nuanced babbling, explained as vowel sounds and vowel-consonant combinations, resulting in a notation in the child’s health records.28 However, the validity of the question, the age for testing, the unstandardised terminology and instructions are all potential shortcomings of the present procedure. At the 10-month visit, the CHS nurse ask about single word understanding, and use of CB, but in a non-standardised way. If instead the standardised babbling questions in the current study were to be used, the CHS nurse could be more confident that the child really did or did not have established CB.

Not having established CB was associated with an increased risk of earlier language disorder according to the language screening 2 years later (PPV 40%). Previously, late CB onset has mainly been associated with a poor expressive vocabulary and articulation.11,13,27 However, the language screenings at 2.5/3 years are multifaceted and represent a broader range of language and communication abilities. A follow-up study is needed to investigate if the children with an early lack of CB represent a subgroup of children with a specific speech production disorder or more general communicative disabilities.

The major shortcoming of the babbling questions was the low sensitivity with only 6 out of 71 children (8%) with later suspected language disorder identified as not having CB at 10 months. Although most children with established CB at 10 months had a typical language development (NPV 93%), most children with a suspected language disorder also had established CB at 10 months. Thus, having CB or not at 10 months cannot, based on our data, be considered to predict screening outcome 2 years later. Unfortunately, we did not perform the babbling assessment among the children with reported CB at 10 months, and we cannot analyse the underlying factors behind the low sensitivity. One possible factor could be that children who have not reached the CB stage by 10 months are missed by the babbling questions, and another that most children with failed later language screening do reach the CB stage by 10 months.

However, the babbling questions did contain some valuable information. Among the 15 children in the non-CB group, slightly less than half the group, 6 children, failed the language screening 2 years later (PPV 40%). This is a greater proportion than the proportion of children with language disorder in general (7.8%).29

| Babbling questions | Language screening |
|--------------------|--------------------|
|                    | Failed screening | Passed screening | Row sum |
| Not CB             | 6               | 9               | 15      |
| Established CB     | 65              | 836             | 901     |
| Column sum         | 71              | 845             | Total: 916 |
Thus, there could be a merit in directing parents of a child without CB at 10 months to an SLP for an assessment if a proactive approach for children at risk should be advocated in favour of a wait-and-see attitude. On the other hand, many children without CB at 10 months but with passed language screening 2 years later (an estimated 60%) would have received the added services without any real need. Thus, the costs and potential harm in such an arrangement must be carefully considered.

The overall predictive validity of the babbling questions, measured by the diagnostic odds ratio (8.57), was on a similar or lower level as other screening instruments for language disorder reviewed by Sim and colleagues. However, when comparing the results of this study with others, we must consider that the babbling questions were applied to younger children and aimed to predict language development over a longer time interval (20–26 months) than other studies. Further, the study was performed in the actual CHS setting and not in an experimentally controlled setting, hence higher ecological validity was expected at the expense of some measurement error and correspondingly, lower predictive accuracy. The generalisability of the study was supported by the comparison to a group of children who did not answer the babbling questions.

Some limitations must also be acknowledged. As mentioned above, the lack of babbling assessment by an SLP for children with established CB at 10 months did not allow us to fully answer why the predictive validity of the babbling questions was poor. Despite no substantial differences between the two screening procedures, and good collaboration with the CHS centres, the data collection had to be individually adapted for each CHS centre which might have affected the number of children dropping out. We only identified a small number of children without CB which led to our estimates of predictive validity being imprecise to some degree, see the confidence intervals in Table 2. Further, our sampling strategy has most likely underestimated the proportion of children without CB in the general population since our sample contained children within the standard CHS, and not only children identified as being at-risk of not developing typical babbling – such as children with hearing difficulties, cleft palate or neurodevelopmental disabilities. Indeed, Oller et al., who sampled from a high-risk population, estimated the proportion to be around 3.1%. However, as we wanted to investigate if babbling status at 10 months could help predict children at risk of later language disorder not previously identified, we believe our sample addresses this research question more accurately.

| Statistic | Value | Interpretation | 95% CI |
|-----------|-------|----------------|--------|
| Sensitivity | 8% | 8% of children with later failed language screening was predicted as such through not having established CB at 10 months | 3%, 17% |
| Specificity | 99% | 99% of children with later passed language screening was predicted as such through having established CB at 10 months | 98%, 99% |
| LR+ | 7.93 | 7.93 times greater probability of not having CB among the children with later failed language screening than among the children who passed the language screening | 2.91, 21.66 |
| LR− | 0.93 | 0.93 times smaller probability of having established CB among the children with later failed language screening, than among the children who passed the language screening | 0.86, 0.99 |
| DOR | 8.57 | 8.57 times greater odds that children with later failed language screening did not have established CB at 10 months, than the same odds among children who passed the language screening | 2.96, 24.83 |
| PPV | 40% | 40% risk of later failed language screening if a child did not have established CB at 10 months | 20%, 65% |
| NPV | 93% | 93% chance of later passed language screening if a child had established CB at 10 months | 92%, 93% |
| RR | 5.54 | 5.54 greater risk of later failed language screening if a child did not have established CB at 10 months, than if the child had established CB at 10 months | 2.86, 10.76 |

Abbreviations: DOR, diagnostic odds ratio; LR+/LR−, positive/negative likelihood ratio; PPV/NPV, positive/negative predictive value; RR, risk ratio.

*PPV and NPV were calculated after taking the proportion of children with failed language screening into account (7.8%).
6 | CONCLUSION

This study suggests that the babbling questions could be included in the 10-month surveillance at the Swedish CHS as valid measures of babbling development, but they cannot predict language screening result at 2.5/3 years.

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CONFLICT OF INTEREST

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SUPPORTING INFORMATION
Additional supporting information can be found online in the Supporting Information section at the end of this article.

APPENDIX 1

Child name or ID: Date: Observer: _________________________________

Babbling questions to parents (In Swedish: Jollerkoll)

1. What sounds does your child make?
Caregiver’s response:

Clear evidence of canonical babbling? YES NO
If NO, please continue to section 2

2. Can you give examples, how does your child sound?
Caregiver’s response:

Clear evidence of canonical babbling? YES NO
If NO, please continue to section 3

3. Does your child make sounds like this: da, baba, dædæ, ma, nana, ga?
Circle your answer YES NO

Comments/examples of sounds or words:

Date and result of a Hearing test (newborn hearing assessment or other):

With a no-answer on all three questions, a CB assessment by a speech and language pathologist is recommended.