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# **DELAYED BABBLING AT 10 MONTHS: OBSERVATION, DETECTION, AND A TWO-YEAR FOLLOW-UP**

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Delayed babbling at 10 months: observation, detection,  
and a two-year follow-up  
THESIS FOR DOCTORAL DEGREE (Ph.D.)

By

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*“Now what, will you spend the rest of your working days doing “dadada” with children?”,  
you asked me a long time ago. Apparently, you were right about that too.*

*To you, mamma Romana.*

## POPULAR SCIENCE SUMMARY

“Ada da da”. The baby looks straight ahead, pointing in the same direction. Daddy looks at the baby, nods, and smiles. “Yes, that’s right”. “Da!”, says the baby again. Daddy goes: “yes, dadada” with a smile. And then he stops, and suddenly starts to laugh: “daddy, did you really say daddy?”.

Combinations of consonants and vowels, like “da” in the example above, sounds like ordinary speech and are the building blocks that eventually will develop into words. These combinations are called canonical babbling (CB) and there is a risk for later language disorder if a child does not use them at 10 months. The Child Health Services (CHS) check that the child reaches important milestones during pre-school years. Therefore, CB would be a suitable target for an early screening at 10 months at the CHS center. The aims of the thesis were to find valid methods to assess the child’s babbling and to examine if a CB screening at 10 months could sort out children who were at risk of developing language disorder at 2.5 or 3 years. We also wanted to see how children who did not use CB at 10 months developed language, cognitive and motor abilities at 12, 18 and 36 months of age. The last research question was if parents used answers in the way the dad in the example above did, that is in ways that support language development.

As a method for assessment, it was found that the observations of babbling, using a systematic observation form, could be trusted. Most children without risk at 2.5 or 3 years passed the CB screening. Many of the children identified at 2.5 or 3 years with risk for language disorder were missed. On the other hand, the CB screening found some of the at-risk children at 2.5 or 3 years as being at risk already at 10 months. For about half of them, the consonant system and spoken vocabulary at 18 months were smaller than in children with typical babbling described in other studies. For one out of five children this limitation remained up to 3 years. It turned out that for these children, the language disorder was part of a more general disability, affecting cognitive and motor abilities as well. The parents to children with delayed CB, did not comment or imitate/expand after their child’s CB utterances as much as they could have done.

In summary, the CB screening dismissed most children without risk for language disorder at 2.5/3 years but found only a few of the children with this risk. This makes the CB screening somewhat problematic. Instead, the information provided by the CB questions (Swe. Jollerboll) can be included in the current surveillance at the 10-month-visit at the CHS center and the observation form can be used for the following specialist assessment for those who need it. In this way, the CHS can take advantage of the specific, predictive details on the child’s babbling and the individual child at-risk can have early contact with a specialist for assessment, intervention, and follow-up. Intervention could be parent-oriented with focus on exploring ways to adjust the answers to the child’s different babbling utterances, just as the dad in the example above did.

## POPULÄRVETENSKAPLIG SAMMANFATTNING

"Aba ba ba". Barnet tittar rakt fram och pekar i samma riktning. Pappa tittar på barnet, nickar och ler. "Ja, det stämmer." "Ba!", säger barnet igen. Pappa säger: "Ja, bababa" med ett leende. Och sen, plötsligt, stannar han upp och börjar skratta: "Pappa, sa du verkligen pappa?".

Kombinationer av konsonanter och vokaler, som "ba" i exemplet ovan, låter som vanligt tal och är byggstenarna som så småningom utvecklas till ord. Dessa kombinationer kallas stavelsejoller och det finns risk för senare språkstörning om ett barn inte använder dem vid 10 månaders ålder. Barnhälsovården (BHV) kontrollerar att barnet når viktiga milstolpar under förskoleåren. Stavelsejoller skulle vara ett lämpligt mål för en tidig screening vid 10 månader vid barnavårdscentralen (BVC). Syftet med avhandlingen var att hitta giltiga metoder för att bedöma barns joller och undersöka om en jollerscreening vid 10 månader kunde sortera ut barn som riskerar att utveckla språkstörning vid 2,5 eller 3 år. För de barn som inte hade stavelsejoller vid 10 mån undersöktes också den fortsatta utvecklingen av språk, kognitiva och motoriska förmågor vid 12, 18 och 36 månaders ålder. Den sista forskningsfrågan var om föräldrar använde svar på joller på det sätt som pappan i exemplet ovan gjorde, det vill säga på ett sätt som stödjer språkutvecklingen.

Som metod för bedömning gick observationer av joller, med hjälp av ett systematiskt observationsformulär, att lita på. De flesta av barnen utan risk vid 2,5 eller 3 år klarade jollerscreeningen. Screeningen missade många av barnen med risk för språkstörning som identifierades vid 2,5 eller 3 år. Å andra sidan fann jollerscreeningen några av de barn som identifierades vid 2,5 eller 3 års ålder som var i riskzonen. För ungefär hälften av dem var konsonantuppsättningen och det expressiva ordförrådet vid 18 månader mindre än hos barn med typiskt joller beskrivna i andra studier. För ett av fem barn var denna begränsning kvar upp till 3 års ålder. Det visade sig att för dessa barn var språkstörningen en del av en mer allmän funktionsnedsättning, som också påverkade kognitiva och motoriska förmågor. Föräldrar till barn med försenat stavelsejoller använde inte kommentarer eller imitationer/expansioner efter sitt barns stavelsejoller lika mycket som de skulle kunna ha gjort.

Sammanfattningsvis kunde jollerscreeningen utesluta de flesta barnen som inte var i riskzonen vid 2,5/3 år, men fann bara en del av barnen som uppvisade denna risk vid 2,5/3 års ålder. Det gör jollerscreeningen något problematisk. I stället kan informationen som frågorna i jollerscreeningen (Jollerroll) ger inkluderas i det nuvarande 10-månaders besöket på BVC och observationsformuläret användas vid efterföljande specialistbedömning för de som behöver. På detta sätt kan de specifika och prediktiva detaljerna om barnets joller tas till vara på BVC och det enskilda barnet i riskzonen kan få tidig kontakt med en specialist för bedömning, intervention och uppföljning. Interventionen kan ha föräldrainriktning med fokus på att upptäcka sätt att anpassa sina svar till barnets joller olika jolleryttranden, precis som pappan i exemplet ovan gjorde.

## ABSTRACT

Babbling variables that can predict later expressive language, such as presence and onset of canonical babbling (CB), presence of anterior stops and consonant inventory, are suitable targets for a CB screening and the Swedish Child Health Services (CHS) is a suitable framework for such a screening. The overall aim of the thesis was to find means for identification and valid care for children with delayed CB and foundations for future intervention. The specific aims were to investigate babbling observation as method for assessment of babbling, the predictive validity of a CB screening at 10 months, follow-up assessments of children who failed the CB screening and parent's use of a responsive approach in interaction with their 10-month-old children who either lacked or used CB.

The thesis consists of five studies, including 1219 participants at 10 months recruited for the project and material for another 149 children aged 9 to 21 months collected for other studies. Children without and with known medical conditions, such as hearing loss, cleft palate, and neurodevelopmental disorders, were included. In study I and II the validity and accuracy of observation as a method for identifying CB was investigated. In study III, the predictive validity of a CB screening was evaluated by comparing the result of a CB screening at 10 months with the result of the language screening in use within the CHS at 2.5/3 years. In study IV, babbling, expressive language, cognitive, and motor abilities in children who failed the CB screening at 10 months were followed-up at 12, 18 and 36 months by babbling observations, parent questionnaires (SECDI) and clinical assessments (SVANTE, Bayley). In study V, parent's responses following two different child utterance types in a clinical group of children were examined, to find a starting point for an intervention for children with delayed CB.

The results showed that babbling observation during interaction with a parent was a valid and reliable method to identify children who used/did not use CB and other predictive consonant variables. Observation of the variables could, to a clinically significant level, separate children in a clinical and a non-clinical group. The CB screening at 10 months had high specificity and negative predictive value. However, the sensitivity was low. For some children who failed the CB screening, expressive language was delayed at 18 months and the delay continued up to 36 months. For one out of five children, the delay in CB was an early sign of a more comprehensive developmental disability. Regarding parent's response types, parents to children with delayed babbling used significantly more acknowledgements following CB than following other vocalizations, which can help children stay in the interaction but does not provide word-shaping models.

The CB screening at 10 months, using valid questions to parents in the first step and valid, reliable, and easy-administered observations of CB variables in the second step, could dismiss most of the children who passed the language screening at 2.5/3 years and identify some of the children who failed the language screening at that age. Although there is a gain in valid and specific information on the babbling of the identified children and benefits of early intervention, the CB screening as a new universal screening within the CHS is problematic. Instead, including the specific CB questions in the current developmental surveillance at the 10-month-visit is suggested, and if needed they can be followed by a specialist assessment.



## SAMMANFATTNING

Jollervariabler som kan förutsäga senare expressivt språk, som start och förekomst av stavelsejoller, förekomst av främre klusiler och konsonantuppsättning, är lämpliga mål för en jollerscreening och svensk barnhälsovård (BHV) är ett lämpligt ramverk för en sådan screening. Det övergripande syftet med avhandlingen var att hitta sätt att identifiera och ta hand om barn med försenat stavelsejoller och en bas för framtida intervention. De specifika målen var att undersöka observation som metod för bedömning av joller, den prediktiva validiteten i en jollerscreening vid 10 månader, uppföljande bedömningar av barn som missat jollerscreeningen och föräldrars följsamma förhållningssätt i interaktion med sina 10 månader gamla barn som antingen saknade eller använde stavelsejoller.

Avhandlingen består av fem studier och omfattade 1219 deltagare vid 10 månaders ålder som rekryterades till detta projekt och material för ytterligare 149 barn i åldern 9 till 21 månader som samlats in från andra studier. Barn utan och med kända medicinska tillstånd, som hörselnedsättning, gomspalt och utvecklingsavvikelse, ingick. I studie I och II undersöktes validitet och noggrannhet i observation som metod för att identifiera stavelsejoller. I studie III utvärderades den prediktiva validiteten i jollerscreeningen genom att jämföra resultatet på jollerscreeningen vid 10 månader med resultatet på den språkscreening som används inom BHV vid 2,5/3 års ålder. I studie IV följdes joller, expressivt språk, kognitiva och motoriska förmågor upp vid 12, 18 och 36 månader hos barn som missat jollerscreeningen. Jollerobservationer, föräldraformulär (SECDI) och kliniska bedömningar (SVANTE, Bayley) användes. I studie V undersöktes föräldrars responser efter två olika typer av barn yttranden i en klinisk grupp av barn för att hitta en utgångspunkt för intervention för barn med försenat stavelsejoller.

Resultaten visade att observation var en giltig och tillförlitlig metod att identifiera barn som använde/inte använde stavelsejoller och andra prediktiva konsonantvariabler. Observation av variablerna kunde skilja ut barn i en klinisk och en icke-klinisk grupp. Jollerscreeningen hade hög specificitet och negativt prediktivt värde. Däremot var sensitiviteten låg. För vissa barn som missat jollerscreeningen var expressivt språk försenat vid 18 månader och förseningen fortsatte upp till 36 månader. För ett av fem barn var förseningen av stavelsejoller ett tidigt tecken på en mer omfattande utvecklingsavvikelse. När det gäller föräldrars svarstyper, använde föräldrar till barn med försenat joller signifikant fler bekräftelser efter stavelsejoller än efter andra vokaliseringar, vilket kan hjälpa barnen att stanna kvar i interaktionen men inte ger någon modell för att forma ord.

Jollerscreeningen vid 10 månader, med valida frågor till föräldrar i ett första steg och valida, tillförlitliga och lättadministrerade observationer av jollervariabler i ett andra steg, kunde avfärda barn som klarade språkscreeningen vid 2,5/3 år och identifiera några av de barn som missade språkscreeningen vid 2,5/3 år. Även om man kan dra nytta av valid och specifik information om joller hos de identifierade barnen och det finns fördelar med tidig intervention, är jollerscreeningen som en ny universell screening inom BHV problematisk. I stället kan tillägg av de specifika frågorna om stavelsejoller i den nuvarande övervakningen av utvecklingen under 10-månadersbesöket rekommenderas, och vid behov kan de följas av en specialistbedömning.



## LIST OF SCIENTIFIC PAPERS

- I. **Lieberman, M.**, & Lohmander, A. (2014). Observation is a valid way of assessing common variables in typical babbling and identifies infants who need further support. *Acta Paediatrica* 103(12), 1251-1257
- II. Lohmander, A., Eriksson, S., Holm, K., & **Lieberman, M.** (2017). Observation method identifies that a lack of canonical babbling can indicate future speech and language problems. *Acta Paediatrica*, 106(6), 935-943
- III. **Lieberman, M.**, Sand, A., Lohmander, A., & Miniscalco, C. Can a simple babbling screening at 10 months within Child Health Services predict language difficulties at 2.5/3 years of age? *Under review, manuscript submitted.*
- IV. **Lieberman, M.**, Hagberg, B., Lohmander, A., & Miniscalco, C. Follow-up of 3-year-old children with delayed babbling at age 10 months. *Manuscript.*
- V. **Lieberman, M.**, Lohmander, A., & Gustavsson, L. (2019). Parents' contingent responses in communication with 10-month-old children in a clinical group with typical or late babbling. *Clinical Linguistics & Phonetics* 33(10-11), 1050-1062.

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## LIST OF ABBREVIATIONS

|                      |   |
|----------------------|---|
| ASD                  | Autism spectrum disorder  |
| CB                   | Canonical Babbling  |
| +CB                  | Positive CB screening   |
| CBR                  | Canonical Babbling Ratio  |
| CBR <sup>UTTER</sup> | Proportion canonical utterances divided by total number of utterances |
| CHS                  | Child Health Services   |
| CP                   | Cleft Lip and Palate  |
| CP                   | Cleft Palate  |
| DLD                  | Developmental Language Disorder                                       |
| +L                   | Positive Language Screening   |
| NPV                  | Negative Predictive Value   |
| OME                  | Otitis Media with Effusion  |
| PCC                  | Percent Consonant Correct   |
| PCC-A                | Percent Consonant Correct, Adjusted for age                           |
| PPV                  | Positive Predictive Value   |
| SECDI                | Swedish Communicative Development Inventory                           |
| SLP                  | Speech and Language Pathologist                                       |
| SVANTE               | Swedish Articulation and Nasality Test                                |
| TD                   | Typical Development, Typically Developing                             |

# 1 INTRODUCTION

Developmental language disorder (DLD) is long lasting and have serious impacts on a child's social, cognitive, and learning functions. DLD can affect both receptive and expressive language (Bishop, Snowling, Thompson, & Greenhalgh, 2016) and the domains being affected can vary during development. Instead of waiting until symptoms are manifested, early identification of children at risk can lead to specialized guidance, earlier diagnosis, and intervention. Even though the DLD condition itself will persist, the benefits of receiving help to facilitate, stimulate and treat the disorder is desirable (Bishop et al., 2016). The Child Health Services (CHS) in Sweden is responsible for thorough surveillance of health issues in children between 0-5 years of age and it reaches almost 100% of the eligible children in the age span (National Board of Health and Welfare, 2014). The regular use of surveillance protocols and screening instruments allows the CHS nurse to build a personal knowledge and experience of the individual child and its family which creates a basis of trust for the family. The CHS has a 50-year long history of including communication and language in the surveillance of child health and well-being, and a major focus is guidance in parenthood under different living circumstances.

A starting point for the present thesis is the babbling screening suggested in the late 90's in the so-called telephone project (Oller, Eilers, Neal, & Lewis, 1998; Oller, Eilers, & Basinger, 2001; Oller, Eilers, Neal, & Schwartz, 1999). In that project, 3400 children in a high-risk population, were contacted by phone with questions to parents on their 10-month-old child's babbling. Children with and without canonical babbling (definition in later sections) were assessed by specialists to confirm the parent's answers and the children were followed-up at 12, 18 and 30 months. To our knowledge, the suggested screening procedure has not been acted out in a primary care setting and the present study aim to evaluate if such a screening could be useful within the Swedish CHS framework.

## **2 EARLY EXPRESSIVE LANGUAGE DEVELOPMENT**

Bloom and Lahey have presented a framework for the language ability which explains how three major aspects, form, content, and use, interact (Lahey, 1988). During development each aspect develops on its own, while at the same time, they interact and affect each other. This holds true for children with DLD as well. Some features might be characteristic for a child at a certain age, while other features are more salient at a later timepoint. This thesis focuses on two of the three aspects, the form and use of early expressive language. The continuity in expressive language develops from the first speech-like utterances, so called protophones during the first months (Oller, 2000) through the milestone of smoothly combining vowel sounds and consonant sounds, the canonical syllables, to eventually reaching the first words. To reach the first word productions, the basis for semantics and future syntax, mastering of the canonical syllables is demanded. For this project the association between babbling variables and later word and consonant production is in focus, whereas later expressive abilities such as syntax and narratives will not be addressed.

### **2.1 BABBLING DEVELOPMENT**

Although ending up in comparable descriptions of babbling development during the first year, models of babbling development differ in included vocalization types, design, and ambient language (Vihman, 2006). Stark (1980) categorized vocalizations matching the earliest stages, such as crying, discomfort/comfort sounds and vegetative sounds (Stark, 1980) and Roug and colleagues focused on early vocalizations in Swedish children (Roug, Landberg, & Lundberg, 1989). The infraphonological model (Oller, 2000) exclude vegetative sounds, highlights protophones (that is, speech-like sounds) and emphasize how vocalizations gradually develop to be more speech-like and thereby, prepares the child for speech. Brief vocalizations, that is phonation on expiratory air is significant during the early phonation stages between 0 to 4 months, at first with airflow alone and later together with simultaneous movements of the lips or tongue. During the expansion phase, at about 4 to 6 months, vowel-like sounds occur with variation in intensity and pitch and combinations of stopped and fluent airflow are tried out (Oller, 2000). Onset of speech-like combinations of consonants and vowels with rapid transition in between, so called canonical babbling (CB) occur between 6 and 8 months for most children and by the age of 10 months at least 93% of all children use CB. A common measure of being in the CB stage is that at least 15% of the utterances are canonical (Oller, Eilers, Steffens, Lynch, & Urbano, 1994; Oller et al., 2001). After onset, the proportion of CB increases with age (Cychosz et al., 2021; Oller et al., 2001), while most utterances consist of other vocalizations for a long period of time (Oller, Ramsay, Bene, Long, & Griebel, 2021).



Furthermore, vocal development, including the CB onset is universal across languages (Oller, 2000). In a recent study of audio recordings in children's daily environment in five culturally and linguistically different settings, there was high consistency in proportion CB syllables in the different settings (Cychosz et al., 2021). The results were consistent with earlier reports (Oller, 2000; Oller et al., 1994), the proportion of 15% CB utterances, the cutoff described as the CB onset (Oller et al., 1994), was reached at the age of seven months on average and for most children before 10 months (Cychosz et al., 2021). The age for onset of CB is robust under different living conditions, for example multi-lingualism and low socio-economic status, and through different potential risk factors, for example prematurity (Nathani, Ertmer, & Stark, 2006; Oller, Eilers, et al., 1998; Törölä, Lehtihalmes, Heikkinen, Olsén, & Yliherva, 2012). Stops are the earliest "true" consonants, that is consonants "involving complete or nearly complete supraglottal closure" (Vihman, 2006, p. 95) and are particularly salient in CB. Accordingly, the most typical consonants are anterior stops, first t/d and then p/b (McCune & Vihman, 2001; Stoel-Gammon, 2011). By 6 months of age, the child uses two to three consonants-like sounds and at 10 months between six to seven (Stoel-Gammon, 2011). After the child has learnt to combine one unique consonant with a vowel (e.g., /ababa/), the next step is to vary the consonant in the same utterance, so called variegated babbling (e.g., /daba/).

Besides preparing for speech, vocalizations also prepare for language use. The flexible functionality is another aspect of early vocalizations that prepare for language use. Protophones could be produced in any situations and in any emotional state, and in this sense, they are functionally flexible. Oller (2000) describes this as a defining property of protophones as opposed to vocalizations with fixed functions, for example laughter or distress sounds. Since the same flexibility is characteristic of words and language, also produced regardless of affect, communicative intention, or situation, protophones prepare for language use and communication. In a recent study, it is argued that this functional flexibility is one of the underlying factors of the great predominance of protophones in the child's early vocalization repertoire (Oller et al., 2021). Protophones was found to be five times more common than fixed functional sounds (i.e., laughter and distress sounds) in everyday recordings across the first year of life. With this overbalance of protophones the child can explore sounds carefully, listen to them, receive sensory input, and handle them in a multimodal way. Children seem to enjoy this vocal play, and repeat it over and over and eventually, when mastering it they start using it for other purposes, i.e., communicating and speaking. The driving force behind this vocal play, used when the child is focusing on a toy or playing by itself, has been described as an intrinsic force within the child, an endogenous motivation (Long et al., 2020). Another possible

motivation that drives babbling development forward is the social one, described in the thesis section Parents' recognition of CB and their adjustments.

### **2.1.1 Typically developing children**

Canonical syllables are the constituents of most words in language, and several studies confirm the continuity of babbling and word acquisition, both regarding the first words and later expressive vocabulary (Vihman & Greenlee, 1987). The almost exact phonetic similarity between syllables in CB and the syllables of the first words has been pointed out as supporting evidence (McCune & Vihman, 2001), for example “da”, one of the most common syllables in babbling. When the same syllable is produced together with pointing, facial expression and eye contact it can connote a word, for example “där” in Swedish (meaning there). In this way a child will not say his first words without first using CB syllables. Time interval between onset of CB and onset of word production (the first five words) was found to be more than 8 months (Oller, Eilers, et al., 1998). Vihman (2006) described CB syllables as the blocks to build words and argued that the more building blocks, that is the more CB syllables, the easier it will be to produce words. High number of different consonants, mostly true consonants, at 12 and 18 months has been described as positively correlated to expressive vocabulary up to 30 months in children with typical development (TD) (McCune & Vihman, 2001).

Other babbling variables that have been suggested as predictive of expressive language are age for CB onset, proportion of CB and consonant production. In the so-called telephone project, time of onset of CB was examined in a largely high-risk population of children (low socio-economic status, low birth weight, exposure to various illnesses and drugs) and followed-up study at 18, 24 and 30 months (Oller et al., 1999). Smaller expressive vocabulary was noted at all ages for children with CB onset after 10 months. Although receptive vocabulary was smaller at 18 months it had recovered already by 24 months, indicating that CB is related to production more than to perception. High prevalence of stop consonants with anterior placement is associated with good articulation skills at 3 and 5 years in typically developing children, as well as children with cleft lip and palate (CLP) (Klintö et al., 2014).

### **2.1.2 Children in clinical groups**

In clinical groups, the same continuity pattern for consonant production as in TD children is present. In children with profound hearing loss aged 10-12 months, the proportion of CB utterances was associated to verbal skills at 30 months and articulation skills at 36 months (Moeller et al., 2007). Both articulation manner and place are relevant. In a group of 30

children with hearing loss related to otitis media with effusion (OME), with and without cleft palate, presence of dental/alveolar stops and number of different true consonants at 10, 12 and 18 months were significantly correlated with articulation proficiency at 36 months in both groups of children (Lohmander, Westberg, Olsson, Tengroth, & Flynn, 2021). Also, in children with cleft palate, the positive association of total number of consonants and number of different consonant types 18 months and articulation at 3 years of age has been described (Klintö et al., 2014). Children with different neurodevelopmental disorders aged 12 to 22 months (e.g., cerebral palsy, Downs syndrome, developmental delay, chromosome deletion syndrome) had significantly lower CB proportion and lower occurrence of dental/alveolar stop than children in a control group (Nyman & Lohmander, 2018). An exception was seen in the group of children with Downs syndrome where there was no significant difference compared to the control group, which confirmed earlier findings (Smith & Oller, 1981). In this group of children, correlation between number of different true consonants was correlated to percentage correct consonants at 36 months, while children with other neurodevelopmental disorders were not (Nyman, Strömbergsson, Lindström, Lohmander, & Miniscalco, 2021). Measures of vocalizations accounted for 25% of the variance in expressive language. A group of children who were later diagnosed with autism spectrum disorder (ASD), showed significantly lower proportion CB and overall lower volubility (i.e., talkativeness) than a group of TD children at 9 to 12 months was measured in home video recordings (Patten et al., 2014). The overall mean effect size of the association between vocalizations and expressive language in children with ASD was significant and strong in a meta-analysis including nine studies (McDaniel, D'Ambrose Slaboch, & Yoder, 2018).

Looking at language disorders in particular, a group of late-talking children, defined as having less than 50 expressive words at 18 months, had lower proportion of CB utterances compared to TD children as well as lower number of different consonants (4.4 compared to 10.5). (Fasolo, Majorano, & D'Odorico, 2008).

It can be concluded that the presence of CB, and a high frequency of oral stop consonants with anterior, especially dental/alveolar placement, and large consonant inventory can be seen as early signs of good progress in a child's language development. Accordingly, lack of these precursors has been shown in children with future speech difficulties.

### **2.1.3 Parents' recognition of CB and their adjustments**

Parents recognize the CB onset of their child reliably (Oller et al., 2001) and it has been suggested that this is an automated process that parents do by intuition. Furthermore, when asked open-ended questions which evoke their own descriptions of the child's vocalizations, the information parents give is reliable. In the telephone project (Oller, Eilers, et al., 1998; Oller et al., 1999) this kind of open-ended questions were used in the interviews with parents. By the parents' descriptions, the interviewer made judgments of presence or absence of CB, which was confirmed in a laboratory session.

Parents adjust their responses to the child utterances (Gros-Louis & Miller, 2018). A responsive utterance is typically a prompt and contingent (conceptually dependent) response to the child's exploring play or communicative acts (Bornstein, Tamis-LeMonda, Hahn, & Haynes, 2008; Tamis-LeMonda, Bornstein, & Baumwell, 2001). A social feedback loop where parents' responsive behavior plays an important role has been suggested to explain what drives language development forward. The child vocalization triggers a parental contingent response, which in turn brings out another child vocalization. On top of this, since parents are sensitive to the level of the child's vocalizations, a CB utterance which is considered more advanced than a vowel vocalization, will more securely trigger a contingent response which in turn increases the likelihood of another advanced CB utterance (Gros-Louis & Miller, 2018). Tamis-LeMonda and colleagues (2001) described how maternal responsiveness with their typically developing children at 9 to 13 months was related to measures of expressive language milestones at 24 months, for example. Onset of the first words, use of 50 expressive words and word combinations.

Enhancing parents' responsiveness, as a prerequisite for the social feedback loop, would be a suitable goal for intervention for 10-month-old infants with suspected language problems. Evidence of the significance of responsiveness in parent-directed interventions for young children with language disorders could also support the choice of intervention goal (Roberts & Kaiser, 2011). A rationale for its position in language intervention is the connection to language growth in children with developmental difficulties. In a group of children with autism spectrum disorders (ASD) parents' responsive behavior predicted expressive vocabulary six months later, after controlling for engagement and volubility (McDuffie & Yoder, 2010). Follow-in comments to the child's focus of attention was a unique predictor of spoken language three years later (Haebig, McDuffie, & Ellis Weismer, 2013). Siller & Sigman (2008) reported that children with ASD and parents who used a higher degree of responsiveness used more advanced language and shared focus of attention at a follow up one year later. Despite this,

only few studies on intervention for children with autism or risk for autism include enough sample size or clear statement of the active treatment ingredients to make an unanimous recommendation on treatment methods (French & Kennedy, 2018).

## **2.2 EXPRESSIVE VOCABULARY DEVELOPMENT**

Since quantitative aspects of expressive vocabulary is one of the outcome measures in the present thesis project, these aspects will be in focus, whereas description of content and semantics will be left out. The first spoken words usually appear at around 12 months and a mean of five words have been reported at 14.5 months in a group of TD children (Oller, Levine, Cobo-Lewis, Eilers, & Pearson, 1998). The child develops around one to three words a week during the first phase of word acquisition and when the vocabulary consists of 20 to 40 words the rate increases to eight words per week (Barrett., 1995). In this early phase, the relationship between expressive and receptive words can be estimated. When the child produces 10 words, he/she understands 100 (Fenson et al., 1993) and when 45 words are produced the child understands four to five times more. When the expressive vocabulary reaches 45 to 50 words, the development of new words increases further (Foster, 1990) and with that size of vocabulary, the development of two-word-phrases begins (Brown, 1973). Fenson et al., (1993) discuss the large individual variation among the 1500 children with TD who were followed over time and found that at 13 months the mean number of words reported by parents was between 8 to 16 words (reported from a word list, MacArthur Communicative Development Inventories). Lee (2011) suggested that the early individual differences in lexicon size continue all through the language development and into literacy development.

## **2.3 CANONICAL BABBLING ASSESSMENT**

As presented in table 1, there are different ways to assess CB with different outcome measures and underlying material. Analysis of CB can be done by transcribing all speech-like utterances, which allows for assessment of the CB proportion as well as description of consonant inventory and calculation of number of different consonants (Lohmander, Olsson, & Flynn, 2011; Oller & Ramsdell, 2006; Willadsen & Albrechtsen, 2006). Assessment of the proportion CB utterances, for example used to decide if the child has reached the CB stage (Oller & Eilers, 1988), can be done in various ways. At large, the assessment measures the relative use of CB production and “non-CB production” and the most common measure is the Canonical Babbling Ratio (CBR). Originally it was calculated by dividing number of CB syllables by total number

of syllables (Oller et al., 1994). A variation is the CBR<sup>UTTER</sup> which has high agreement with the original CBR based on syllables (Nyman & Lohmander, 2018). In CBR<sup>UTTER</sup>, number of canonical utterances is divided by total number of utterances.

Transcription is a time-consuming method which requires much training and experience from the transcribers. In addition, it has been argued that phonetic transcription overestimates the inventory of sounds (Ramsdell, Oller, Buder, Ethington, & Chorna, 2012). As an alternative method to measure consonant inventory, marking consonant sounds heard at least twice during a recording from a consonant chart can be used. Lately, noting syllables heard by memory after observation (live or from recording), so called naturalistic listening in real time has also been used (Ramsdell et al., 2012; Willadsen, Persson, Patrick, Lohmander, & Oller, 2020).

Outcome measures vary but include place and manner of articulation, presence of anterior versus posterior consonants, presence of stops, nasals, and approximants respectively, consonant inventory and number of different consonant sounds. A variation is to include true consonants (Vihman, 2006). Consonants are “true” in the sense that they cannot stand alone in a syllable and cannot be confused with semi-vowels. Accordingly, number of different true consonants, that is all consonants except glides and glottals have also been reported in studies.

A recent approach in CB assessment is to consider the recording setting, which can be at a clinic or laboratory, or a more naturalistic settings, for example at home. The LENA equipment has made it possible to record all sounds coming from the child and its surroundings (McDaniel, Yoder, Estes, & Rogers, 2020).

Table 1. Overview of CB assessments, outcome measures and underlying material for assessment.

|                            | <b>Outcome measures</b>   | <b>Material</b>            |
|----------------------------|---|----------------------------|
| <b>CB onset</b>            | Binary outcome, yes/no  | Observation forms          |
|                            | CBR measures  | Transcription, coding      |
| <b>CB proportion</b>       | CBR measures, for example CBR <sup>UTTER</sup>                              | Transcription or coding    |
| <b>Consonant inventory</b> | Place and manner categories of articulation, binary outcome presence or not | Observation forms          |
|                            | Number of different consonant sounds  | Transcription or notations |
|                            | Number of different true consonant sounds                                   | Transcription or notations |
| <b>Setting</b>             | Laboratory/clinic   |                            |
|                            | Naturalistic everyday setting   |                            |

## 2.4 SCREENING AND SURVEILLANCE

Early detection of cases with a suspected disease or medical condition can be done in different ways. Screening can be defined as a “rough sorting process...separating the people who probably do have the condition from those who probably do not (WHO, Regional Europe, 2020, p. 3). It does not provide certainty or any diagnosis, but a probability that the individual is at risk or not by separating individuals into two different groups: probably healthy and probably sick. Screening can be used on the whole population or in selected groups, focusing on individuals without current symptoms, which makes it different from other examinations that aim at diagnosing people with symptoms. The WHO published a list of 10 principles for screening programs (Wilson & Jungner, 1968), which has been summarized in four main areas: 1) the significance of the condition/illness, 2) the validity of the screening method 3) available effective treatment methods and 4) the cost-effect relationship (Swedish agency for health and technology assessment and social services, 1996). The 10-principle-list is also a foundation for implementation and evaluating screening programs in Sweden (National Board of Health and Welfare, 2019).

Another way of detection of cases with suspected medical conditions is surveillance, which can be defined as “close and continuous observation” (Wilson & Jungner, 1968) and conveys a sense of an on-going process of recognizing who may be at risk for a condition. There is an individual focus, and it is usually performed repeatedly at certain time intervals. It differs from other medical examinations as it does not start with a symptom or injury, but a pre-planned focus decided by the health system. How systematic or standardized the method of surveillance is can vary.

Considering DLD, early detection means recognizing a child’s risk of DLD early in life and before the condition is apparent (the Bercow Report, 2008). The purpose is to offer intervention and efforts to decrease the consequences of the probable disorder. Since a secure DLD diagnosis is difficult to make under the age of 3 years (Bishop et al., 2017), it would be unfortunate for the child with probable risk to wait for a diagnosis to receive intervention. Hence, early detection is of special importance. Despite of this, there is no international consensus on implementation of universal screening programs for language disorders during preschool years, and there is more than one reason for this. For example, studies do not clearly state predictive validity and psychometric properties, and they use different tests as golden standard without using standardized cutoffs. There is limited evidence of what is effective in treatment methods and a lack of comparisons between intervention straight after identification and intervention after diagnosis (Siu, 2015, Wallace et al., 2016; Sim et al., 2019; Julien, 2021).

This was confirmed in a recent scoping review (Sansavini et al., 2021), where early predictors of DLD, optimal age range for screening and effective diagnostic tools were discussed. In the included 37 studies, the indirect indication for age recommendation on screening was between 2 and 3 years of age for a diagnosis around 4 years. The American Academy of Pediatrics (Lipkin et al., 2020), promoted a universal model of surveillance and screening of developmental conditions, including language disorders/DLD. A system used over time was recommended to enhance the precision of identifying children at risk compared to relying on professionals' judgement or parents' concern only. This supports the recommendation of continuous formal monitoring of language between ages 2 and 5 years, by Law and colleagues (2017) and the Bercow report in Great Britain (2008). Multiple occasions of surveillance need to take the course of language development into account and create different standards at different milestones.

In conclusion, there is a need for repeated occasions of identification of children with higher risk of DLD during pre-school years, with different methods to match the language milestones and to offer interventions that enhance language development and can "... lead to better health outcomes for some of the screened individuals" (WHO, Regional Europe, p.3)

## **2.5 THE CHILD HEALTH SERVICES IN SWEDEN**

Surveillance of health and development in infants and preschool children has a long history in Sweden. In 1937, by a governmental decision, general services at CHS-centers were set up, open and voluntary for all families and free of charge (Magnusson, Blennow, Hagelin, & Sundelin, 2016). The program has changed over time, orientation towards health surveillance was introduced in the 60's and focus on parenting and promoting child development was emphasized in the 70's. In 1991, national guidelines to regulate the CHS work were published by the National Board of Health and Welfare. The guidelines, updated in 2014, are meant to form the knowledge basis and to contribute to equivalent CHS over the country (National Board of Health and Welfare, 2014). In addition to the national guidelines, there are other sources for method and knowledge support, one is The National Child Health Care Program (swe: Rikshandboken för barnhälsovård) which in detail describe the content, methods, and knowledge foundation of the CHS work (The National Child Health Care program, 2019). At the regional level there is a third set of guidelines that gives direct and detailed information for the specific region (Guide for health care providers, Region Stockholm, 2021; Health Care providers' web, Region Västra Götaland, 2021). Furthermore, the work of the CHS builds upon evidence-based practices which makes demands of quality control, evaluation, and research



(Magnusson et al., 2016). This ambition leads to continuous adjustments and development to validate the national program and to match the contemporary needs of families with young children. With connection to the area of language development, there are on-going projects to improve methods to support language stimulations, for example in co-operation with other society actors (regional libraries, SLP clinics).

Other factors that characterize the Swedish society when it comes to young children and their families is the possibilities of long parental leave during the child's first years and the open day care (in Swedish: öppen förskola), free of charge and open for all. Pre-school for children aged 1 to 5 years, where 85% of all children in the age span, is also open for all and is economically subsidized by the government (Swedish National Agency for Education, 2021).

### **2.5.1 Surveillance of language and communication within the CHS**

The language development is followed continuously with surveillance or formal screening procedures at certain key ages. The efforts, as described in the national guidelines (The National Child Health Care program, 2019), are structured and detailed (see table 2) and include questions to parents as well as direct observations of the child. In general, interaction and communication is discussed at all visits and opportunities for the parents to raise questions or concerns are integrated in the visits. At six months the language surveillance includes nuanced babbling (in Swedish: nyanserat joller), explained as vowel sounds and vowel-consonant combinations (The National Child Health Care program, 2019). The right linguistic term for the mentioned combinations would be canonical babbling (in Swedish: stavelsejoller) and as discussed in earlier sections of the thesis, the debut of CB is often round six to eight months. On the other hand, if CB onset is the target in question and the purpose is identifying risk factors, 10 months of age is a suitable age for including CB (Oller et al., 2001). According to the guidelines, the developmental surveillance at 10 months includes understanding of single words (by direct observation) and use of CB (in Swedish, stavelsejoller), exemplified as “äjä, nana, ba” (The National Child Health Care program, 2019). In the child health care journal, nuanced babbling at 6 months and understanding of instructions at 10 months should be noted with reference to method in use and result (Magnusson et al., 2016).

Table 2. Language surveillance during health visits at key ages (examinations of psychomotor development). (J) means a notation in the CHS journal (Magnusson et al., 2016)

| <b>Age</b>                     | <b>Language standards (and connected standards in the CHS journal)</b>   | <b>What to do if the child does not meet standards</b>   |
|--------------------------------|--|--|
| <b>6–8 weeks</b>               | Response smile (J)<br>Response sounds (J)  | Exclude visual, hearing or interaction difficulties. Exclude parental depression<br>Discuss in the CHS team                        |
| <b>3–5 months</b>              | Follow-up on response smile and sounds<br>Varied vowels  | As 6–8 weeks   |
| <b>6 months &amp; 8 months</b> | Nuanced babbling* (J)<br>Show interest in interaction<br>Looks for hidden toy  | Discuss in the CHS team<br>Consult a medical doctor or psychologist<br>Consider referral to hearing control                        |
| <b>10 months</b>               | Understands single words (J)<br>Plays peek-a-boo (J)<br>Imitates block game (J)<br>Follows instructions<br>Joint attention                           | Follow-up<br>Discuss in the CHS team<br>Consider referring to SLP, psychologist or medical doctor                                  |
| <b>18 months</b>               | Speaks 8-10 words (J)<br>Understands more than 8-10 words (J)<br>Understand instructions (J)<br>Points body parts on demand (J)<br>Find a hidden toy | Guiding/Counselling on language development<br>Consult the CHS team<br>Consider referring to psychologist, SLP or a medical doctor |

\*(Swedish: nyanserat joller)

There are two evidence-based screening programs in use today (The National Child Health Care program, 2019) and both aim at identifying children with suspected DLD at 2.5 (Miniscalco Mattsson, Mårild, & Pehrsson, 2001) or 3 years of age (Westerlund & Sundelin, 2000). Both methods include questions to parents and direct observation. In the 2.5 year screening the direct observation is based on structured play with nine toys. The purpose is to assess the child's ability to comprehend single words and multiword sentences, imitate, name, and use two-word sentences. Miniscalco Mattson and colleagues (2001) performed an evaluation of the screening program where the prevalence was calculated to 10.3% and the method showed high specificity (0.93) and sensitivity (0.69). Out of 100 children with a positive screening result, 87 children received a DLD diagnosis after a detailed assessment by a SLP (Schachinger et al., 2018). In a follow-up study, (Miniscalco, Nygren, Hagberg, Kadesjö, & Gillberg, 2006; Miniscalco, Westerlund, & Lohmander, 2005) showed that children screened positive at age 2.5 years were at high risk of having persistent DLD at age 6 years affecting all

language domains (phonology, syntax, semantics, and pragmatics). When the positive screened children were 7 to 8 years old 60 to 70% had a neuropsychiatric diagnosis (ASD, ADHD or learning problems) alongside language disorder (Miniscalco, Hagberg, Kadesjö, Westerlund, & Gillberg, 2007), now presented as problems with narrating, non-word-repetition, and early literacy (Miniscalco & Dahlgren Sandberg, 2010). The results showing language problems as a risk marker for both persistent language problems in early school years as well as other neurodevelopmental problems have been confirmed in a recent study by the research group (Miniscalco et al., 2018).

The direct observation in the 3-year screening procedure is based on five pictures of everyday objects. The purpose is to assess the child's ability to comprehend sentences, use three-word sentences and ability to cooperate (Westerlund, Bergkvist, Lagerberg, & Sundelin, 2002; Westerlund & Sundelin, 2000). At a thorough check of SLP journals one year after, 30% of the children with positive screening showed severe difficulties and 40% were assessed as having moderate difficulties (Westerlund et al., 2002). Almost two thirds, (61%) of the positively screened children in the 3-year screening were enrolled in habilitation services for children with neurodevelopmental disorders in school-aged years, for example with learning disabilities and neurodevelopmental difficulties (Westerlund et al., 2002). A modified version of the 3-year screening for use six months earlier have been proposed. The adaptation of the screening instrument included a relaxation of the production criteria of three-word sentences to two-word sentences (Nayeb et al., 2019). The adapted version used at 2.5 years identified children with DLD with satisfactory accuracy. For bilingual children, four models for screening procedure have been tried with the modified screening at 2.5 years (Nayeb, Lagerberg, Sarkadi, Salameh, & Eriksson, 2021). The model based on direct screening at 2.5 years on the child's both languages attained the most adequate accuracy and thereby, it was recommended for use.

There has been attempts to screen children below age 2.5-3 years for language disorder in Sweden, for example by parent-reported vocabulary at 18 months (Westerlund, Eriksson, & Berglund, 2004). There were too many of the identified children who developed typical language by 3 years of age (Westerlund et al., 2004) and parent-reported vocabulary was not recommended as a screening tool. Another study of a screening tool at 18 months included observations of play behavior, interaction, expressive and impressive skills by a specialist (SLP student) (Bruce, Kornfalt, Radeborg, Hansson, & Nettelbladt, 2003). There was a significant positive correlation between the evaluations at 18 months and results on formal language tests at 4.5 years of age. In a recent study of the psychometric properties of a general parent questionnaire on communication and language (the Infant Toddler Checklist) given at the 18-

month-visit, suggested that the questionnaire could be used to identify children at 18 months in need of intervention to enhance communication (Fäldt et al., 2021).

An attempt to identify children under age 12 months at risk for atypical development have been carried out. Systematic observations of social interaction, communication, and motor skills by the CHS nurse were used as a screening instrument at 8 months in a primary care setting (Sivberg, Lundqvist, Johanson, Nordström, & Persson, 2016). The observational instrument was concluded to be a useful tool to for the CHS nurse to observe and identify children who would benefit from care and intervention.

## **2.6 SUMMARY**

Theoretically, CB is a relevant target for early identification of children with DLD, with the purpose of initiating follow-up assessments and intervention for children with risk for expressive language disorder. Surveillance and screening should identify children who are exposed to high risk for the condition which also seem to match CB as target for an early screening. Primarily, this builds on the fact that CB constitutes precursors to spoken words (Vihman and Greenlee, 1987) and that late onset of CB is correlated to developmental difficulties and expressive language difficulties later (Oller et al., 1998). An early screening of CB was suggested at 10 months (Oller et al., 1999) but, to our knowledge, has not been tested in a study. As described by Oller and colleagues (2001), parents can reliably recognize onset of CB with a few standardized questions which could constitute an easy-administered cost-effective way to identify children with late babbling within the CHS framework.

## **3 RESEARCH AIMS**

### **3.1 GENERAL AIMS**

The overall aim was to examine if a simple CB screening within the CHS framework could identify 10-month-old children with risk of developing expressive language difficulties two years later. A second aim was to examine how parents of children at this age with a known risk for future speech and language difficulties, respond to the child's different utterance types. The purpose was to find means for valid care and foundations for future intervention for children with delayed CB.

### **3.2 SPECIFIC AIMS**

The specific aims were to:

- investigate the validity and diagnostic accuracy of systematic observation of CB variables that are predictive of future expressive language development by using a babbling observation form (study I and II)
- evaluate if a simple CB screening at 10 months at the CHS center can identify children who fail the later language screening within the CHS (Study III)
- investigate how speech, language, cognitive and motor ability develop in children identified as having delayed babbling, according to a CB screening at 10 months (study IV)
- examine parents' verbal contingent responses to the vocalization utterances and CB utterances of their 10-month-old children at risk for speech and language difficulties (study V)

## 4 MATERIALS AND METHODS

### 4.1 PARTICIPANTS AND RECRUITMENT

In all, this thesis include data from 1403 children. The participating children were between 10 and 36 months and displayed a variety of clinical conditions with risk for speech and language development as well as typical development. An overview of number, ages, and clinical status of the participants in each of the five studies is presented in table 3. Criteria to be included in all studies in the project was that at least one parent spoke native Swedish with the child. Material collected in previous studies allowed for creating new data used in study I, II and V whereas participants in study III and IV were recruited for this project.

Table 3. Table of number and ages of the participants, and their clinical status in study I to V

|                  | No. participants | Child ages    | Clinical status   |
|------------------|------------------|---------------|---|
| <b>Study I</b>   | 38*              | 12, 18 months | Typical development, cleft lip and palate   |
| <b>Study II</b>  | 89**             | 9–21 months   | Typical development, neurodevelopmental disorders, hearing impairment, cleft lip and palate |
| <b>Study III</b> | 1219             | 10–36 months  | Typical babbling development, delayed babbling development                                  |
| <b>Study IV</b>  | 35               | 12–36 months  | Delayed babbling development, delayed language development                                  |
| <b>Study V</b>   | 22               | 10 months     | Typical or delayed babbling development, in a clinical group of children                    |

\*Material (transcriptions and observation forms) collected in earlier studies

\*\*30 children with typical development recruited for this project, remaining material collected in earlier studies

#### 4.1.1 Study I

In total, material from 38 children with and without cleft palate were used, 20 children at 12 and 18 months with typical development and 18 children with unilateral cleft lip and palate (UCLP) at 18 months of which 9 children also participated at 12 months. The material for the children with cleft palate (in total, 18 children) had been collected in a subproject of the international ScandCleft project, aiming at examining the early babbling and speech development in Swedish infants with and without cleft lip and palate (Lohmander et al., 2011). The children without cleft palate (20 children) had been recruited from CHS centers in the same region as the children with cleft palate and were considered as a control group of children.

### **4.1.2 Study II**

In total, there were 89 children in the study, divided into a clinical group aged 9 to 21 months (n=38) and one non-clinical group consisting of children aged 10 months (n=30) and 12 months (n=21). The clinical groups were recruited from Karolinska University hospital and from habilitation centers in the Stockholm area for other studies (Lohmander et al., 2021; Löfkvist et al., 2019; Nyman & Lohmander, 2018) and consisted of eight children born with cleft palate, 20 children with hearing impairment of different background (OME, sensory hearing loss and cochlear implant) and 10 children with neurodevelopmental disorders (such as cerebral palsy, suspected developmental delay or chromosome deletion syndrome). The non-clinical group consisting of 30 children aged 10 months, were recruited for this study from CHS centers in the Stockholm area (Holm & Eriksson, 2013) and 21 children, aged 12 months, recruited from CHS centers in Gothenburg for another study of babbling (Lohmander et al., 2011) with the same project procedure. Hearing in the non-clinical group was normal, as assessed with the newborn hearing screening for the 10-month-old children or a hearing test for the 12-month-old children ( $\leq 20$  dBHL).

### **4.1.3 Study III and study IV**

Data collection was carried out from 2016 to 2019 and it was preceded by a period of preparations together with the CHS centers in the two largest cities in Sweden, Stockholm, and Gothenburg. Information about the project was spread through CHS newsletters and large-scale lectures. Twenty-five CHS centers volunteered to participate, they were equally distributed in the two city regions. Information meetings were held to prepare the CHS nurses on how to inform parents about the project, perform the CB screening and document the results. Parents of 2200 children consented to receive detailed written project information, after receiving oral information by the child's responsible CHS nurse at the ordinary 8-month-visit. Consent forms, signed by both parents, were collected at the 10-month-visit. In total, 1126 children participated in the CB screening at 10 months, which formed the basis for study III and IV. In study III another 93 children (Karlsson & Moretta, 2019) participated by sharing information on the result of their regular language screening (L screening) at age 2.5/3 years, resulting in total 1219 participants. There were 15 children who failed the CB screening and 71 who failed the regular language screening at 2.5/3 years.

In study IV, all 15 children with positive CB screening participated and 21 (31%) out of the 71 children with positive language screening at 2.5/3 years within the CHS, in total 35 participants.

#### 4.1.4 Study V

The material in study V, consisting of video recordings of play sessions between child and parent, was collected within another project aiming at investigating babbling and early speech in children with OME with and without cleft palate (Lohmander et al., 2021). Recordings of 22 children were included in the present study, 15 boys and seven girls. Originally, there were 29 recordings eligible, but seven recordings were excluded due to simultaneous interaction of two parents (n=6) and authors not knowing the language at use during the recording (n=1). The mean age was 9.9 months (range 8.4–11.8). The study group was divided into two subgroups according to their child's babbling level. Fifteen children had >15% CB utterances and were considered to have typical babbling (Oller et al., 1994), whereas seven children had  $\leq$  15% and were considered as having late CB.

## 4.2 ASSESSMENTS AND PROCEDURE

The CB screening in study III was performed by the CHS nurse at the regular 10-month-visit and it consisted of one to three separate questions on the child's babbling directed to the parent (Lohmander, Holm, Eriksson, & Lieberman, 2017; Oller, Eilers, et al., 1998). The questions were: 1) *What sounds does your child make?* 2) *Can you give examples; how does your child sound?* 3) *Does your child make sounds such as, da, baba, ma, nana, ga?* The CHS nurse proceeded to the next question only if the parent failed to give examples of CB and then evaluated each answer as CB or not CB by filling in a screening form.

Babbling assessment from observation at 10, 12 and 18 months in all studies emanated from audio and video recordings of interactions between the child and a parent and the recordings were approximately 30-45 min. Except for in study V, the role of the parent was to act as a partner in the interaction and their speech and acts were not noted. The same instruction, namely, to play and talk as they normally do with the child at home, was given to parents in all five studies and all play situations centered around a fixed set of age-appropriate toys (cuddly toys, cutlery, a boat, a train, a flip book). Most recordings were made at a SLP department at a university hospital (study I, II, part of study III and IV), in a speech lab at a university (study V) and in the local CHS center (parts of study III, IV).

The structured CB assessment from the observations in study I, II, III and IV followed the same procedure but the observation form itself was changed during the project time (described in the Methods section). The SLP noted estimated amount of CB on a visual analogue scale (and of vocalizations in study I), presence of the CB variables and the consonant inventory. The CB variables in study I were high pressure consonants/oral stops, anterior consonants, and oral



dental/alveolar consonants, fricatives, posterior consonants, and glottal placement (yes or no) and in study II, III and IV the three CB variables were oral stops, anterior stops, and dental/alveolar stops. Consonant inventory was assessed by marking of the consonants heard at least twice from a consonant chart.

To document expressive vocabulary, the Swedish version of the MacArthur-Bates Communicative Developmental Inventory (MCDI) (Fenson et al., 1993) was used in study IV (SECDI, words & gestures and words & sentences) (Berglund & Eriksson, 2000; Eriksson & Berglund, 1999). Parents marked the words in a word list that they had heard their child use spontaneously.

To assess consonant production, the Swedish Articulation and Nasality Test (SVANTE) (Lohmander, Lundeborg, & Persson, 2017) was performed. The part of the test that was used consisted of 64 single words elicited by picture naming.

To assess general development The Bayley Scales of Infant and Toddler development III (Bayley-III) (Bayley, 2006) was performed at 12 and 36 months (study IV). The test includes the areas of cognition, language (receptive and expressive subtests) and motor abilities (fine and gross motor subtests).

The parents were given the possibility to freely comment their child's development at all assessment visits (Study IV). This additional informal information was given the same code as other material for each child, and it was saved with the other material.

## **4.3 ANALYSIS**

### **4.3.1 Measures of babbling, consonant production, and expressive vocabulary**

In the CB screening (Study III), judgement of CB (has/has not) was noted on a CB screening form by the CHS nurse, based on the parent's answers on the three questions (Lohmander, Holm, et al., 2017). When "has CB" was noted for any of the three questions, the child was considered to have passed the screening (i.e., negative CB-screening, typical CB). Children with a "has not CB" notation on all three questions were considered as having failed the screening (i.e., positive CB-screening, delayed CB) and were therefore scheduled for the CB-assessment.

Phonetic transcriptions of all utterances were used in study I, with the target of 100 consecutive speech-like utterances. CB was noted as present when 20% or more of the total number of utterances were canonical (Oller & Eilers, 1988). The consonant variables were noted when heard twice or more. In study I and II, the child utterances were also counted and divided in vocalizations or canonical utterances. CB was noted when 15% or more were canonical (Oller et al., 1994). The observation form used in the CB assessments in the study I, II and IV was under development during the project period, hence the variables differed in the studies (table 4).

Table 4. The studied babbling variables by observation in study I, II and V (mo=months)

|              | <b>Study I</b>                             | <b>Study II</b>                   | <b>Study IV</b><br>Observation form,<br>vers 14.3 |
|--------------|--|-----------------------------------|---|
| Presence of: |  |                                   |   |
|              | CB (12 mo)                                 | CB                                | CB (12 mo)  |
|              | Anterior consonants<br>(12 mo, 18 mo)      | Anterior placement                | Anterior stops<br>(12 mo, 18 mo)                  |
|              | High-pressure consonants<br>(12 mo)        |                                   |   |
|              | Oral dental/alveolar consonants<br>(18 mo) | Dental/alveolar stops             | Dental/alveolar stops<br>(12 mo, 18 mo)           |
|              | Oral stop consonants<br>(18 mo)            | Oral stops                        | Oral stops<br>(12 mo, 18 mo)                      |
|              | Fricatives<br>(18 mo)                      |                                   |   |
|              | Posterior oral<br>& Glottal consonants     |                                   |   |
| Notation of: |  |                                   |   |
|              | Consonants heard twice or more             | Consonants heard<br>twice or more | Consonants heard at<br>least twice or more        |

At 36 months (study IV), the consonant production was assessed (SVANTE). The test covers all Swedish consonants, each consonant can be elicited in more than one word and in different word positions. Ten target consonants, that is, the six oral stops /p, b, t, d, k, g/, three fricatives /f, s, ʃ/, and one nasal consonant /n/, are recommended for analysis (Lohmander, Lundeborg, et al., 2017). The elicited words were transcribed with broad whole-word phonetic transcription (Howard & Heselwood, 2002) from the audio-video recordings. For the target consonants, the percentage consonants correct adjusted for age (PCC-A) was calculated by dividing the number of correctly produced target consonants by the total number of elicited target consonants (Klintö et al., 2014; Shriberg, 1993). A consonant was considered established if it was produced at least 50% of the possible elicited times. Mean number of established consonants among the

10 target consonants was calculated. Measures of expressive vocabulary (study IV) was mean number of reported expressive words from the SECDI questionnaire at 18 and 36 months (Berglund & Eriksson, 2000; Eriksson & Berglund, 1999).

#### **4.3.2 Measures of parental contingent responses**

In study V, child utterance and parental response were categorized and annotated in ELAN, version 5.2 a computer software system for multimodal complex annotation of video and audio recorded material (ELAN, 2018). The child utterance types were vocalizations and CB. The categories for parental response types were acknowledgement, comment, imitation/expansion, and directive. Proportion contingent responses after the child utterances were counted as well as proportion of each response type, first for all child utterances together and then divided by the two groups typical babbling and delayed babbling.

#### **4.3.3 Measures of general development**

The Bayley III (study IV) contains three domains, namely cognitive, language and motor domain. The language domain contains a composite of receptive and expressive subtests, and the motor domain a composite of gross motor and fine motor subtests. Raw scores of the subtests were combined and the composite scores were converted into index scores for each child according to the test manual (Bayley, 2006). The mean of the index scores for the +CB screened group and the +L screened group were calculated.

#### **4.3.4 Reliability**

An overview of reliability calculations in the studies are presented in table 5. For presence of the CB variables, percentage agreement, point-by-point, was used for reliability calculation and for number of different consonants and true different consonants (study II and IV) the intraclass correlation coefficient was used. The basis for all calculations were 27 to 29% re-observations by the same observer and 30 to 40% re-observations by another observer in the different studies. Intra-observer agreement varied between 80 and 100% and inter-observer agreement between 80 and 83%. However, in parts of the clinical group in study II (the neurodevelopmental disorder subgroup), the inter-observer reliability was 58% and intra-agreement for one observer was 74%. In terms of parental contingent responses (study V), four recordings (18%) were used for the two annotators for training and calibration. Average inter-rater reliability for parental contingent response types is presented in table 5.

Table 5. Overview of reliability in study I, II, IV and V

|  | <b>Intra-rater reliability</b> | <b>Inter-rater reliability</b> |
|--|--------------------------------|--------------------------------|
| <b>Study I</b><br>Observations   | 100% agreement                 | 100% agreement                 |
| <b>Study II</b><br>Presence CB variables   | >80% agreement<br>>0.83 ICC*   | >80% agreement<br>>0.83 ICC    |
| <b>Study IV</b><br>Presence CB variables 12 mo<br>18 mo<br>Transcriptions, target consonants in words<br>36 mo | 100% agreement<br>100%<br>92%  | 83% agreement<br>95%           |
| <b>Study V</b><br>Contingent responses   | 90,5%                          | 84%                            |

\*except for number of different consonants in the group with neurodevelopmental disorders

#### 4.3.5 Statistical analysis

Descriptive statistics and non-parametric tests were used as the data was not normally distributed. All analyzes were performed using IBM SPSS (versions 25 and 27) for Windows 18.0 software [Armonk, NY].

Sensitivity, specificity, positive and negative predictive value (PPV and NPV) were calculated to compare precisions of the two methods measuring the CB variables in study I and the predictive validity of the CB screening in study III. Sensitivity, specificity, and likelihood ratios were also used in study II as well as odds ratios.

Descriptive statistics were used to calculate prevalence of CB (study II and III) and for CB variables (study IV). Mann-Whitney U test and Kruskal-Wallis were used to test differences between groups and subgroups for number of different true consonants, PCC-A, expressive vocabulary, and Bayley composite scores (Study IV) and for parental contingent responses (study V). The parametric independent t-test was used to test the differences in CBR (Study II) and Wilcoxon signed-rank test in proportion parental response types between the respective groups (Study V). Correlations between outcome measures at different ages were calculated using Spearman's rank correlation (study IV) and univariate regression and binary logistic regression (Study II).

#### **4.4 ETHICAL CONSIDERATIONS**

Ethical approvals were obtained from the Regional Ethical Committee in Stockholm (study II: Dnr 2012/2213-31, Dnr 2013/1989-32, study III, IV: Dnr 015/1401-31, Dnr 2016/267-32, Dnr 2018/1772-32, study V: Dnr 2012/46–32-2). For study I, ethical approval was not needed since it was based on fully anonymized observation forms and transcriptions. For all children both parents gave their written consent to participate.

In the screening and follow-up study (study III, IV) the benefit and risk relationship were considered, and the benefits was regarded to overweight the risks. For the participants there was nothing in the procedure that did include any risks for the child. The procedure was performed during play together with a parent using toys and books or testing with playful and motivating materials, all the time with the parent present and active in the room. If the child showed signs of distress or discomfort the session was paused or ended. There is always a risk of inducing distress or guilt in parents by identifying difficulties or risk factors. To make this risk as small as possible, the test leader answered any question parents might have and made sure that the parents could be in contact with the test leader at any time during the project. The local CHS centers were also familiar with the project and were prepared to answer questions that might arise on the child's development. Another ethical question was the justification of trying to identify risk factors for symptoms that will occur a long time after identification and even if identified early, symptoms that cannot be eliminated. The availability of intervention that can reduce symptoms and parental stress and anxiety can balance this question. The benefits of reaching new knowledge on early babbling and speech development, based on assessments in a Swedish population, will make evidence-based, detailed, and precise guidance to parents possible and create a foundation for developing support programs.



## **5 RESULTS**

### **5.1 STUDY I**

Two different methods, observation and transcription, for assessing babbling variables were compared to examine the validity of a structured babbling observation method. Material from 38 children were used. There was excellent agreement (100%) between observation and transcription for presence of CB at 12 months and anterior consonants at 12 and 18 months. Furthermore, there was high agreement (90%) for high pressure consonants at 12 months and oral stop consonants and dental/alveolar consonants at 18 months (87% and 79% respectively). Observation of these predictive variables had high specificity (range 0.90 to 1) and negative predictive value (range 0.9 to 1) meaning that the observation method could dismiss children who used the variables and that there were few misjudgments (false negatives). Agreement for the variable total number of consonant types (i.e., different consonants) at 18 months was low (24%). However, dividing into groups of 0-5 consonants, 6-10 consonants and more than 10 consonants, the agreement increased to 63%.

### **5.2 STUDY II**

The diagnostic accuracy of the CB observations of 89 children (a clinical and a non-clinical group) was good, the overall percentage of correctly predicted outcomes was 90%. The specificity was high (0.93) indicating that the CB observation could acquit the 10-month-old children with CB and the sensitivity was also high (0.89) indicating that the observations could identify children without CB. The area under the curve showed good discriminative validity (0.832). The accuracy in dividing the children in a clinical and non-clinical group according to presence/absence of the observed babbling variables (oral stops, anterior placement, and dental/alveolar stops) was 60 to 65% with high specificity (0.93-0.97) but lower sensitivity (0.34-0.42), for all variables. The increased risk (in terms of odd ratios) of being in a clinical group for children who lack CB, were high (>10) and statistically significant for all variables.

### **5.3 STUDY III**

Out of the 1219 participating children, 1126 took part in the CB screening at 10 months. Among them there were 16 children with a positive CB screening, indicating a prevalence of 1.4% children with delayed CB. After one child had declined further participation, 12 (80%) of the remaining 15 children were confirmed as having delayed babbling in the CB assessment. At 36 months, 916 (81%) of the 1126 children participated in the language screening (L screening). Predictive validity is presented in table 6. The specificity and negative predictive value (NPV) were high (99% and 93%) which implied that the CB screening correctly rejected

most children who passed the regular L-screening at 2.5/3 years and that children did not risk failing the L screening after a negative CB screening result. However, the sensitivity was very low (8%), indicating that the CB screening could identify only a few of the children who failed the L screening, thereby were at-risk for DLD at 2.5/3 years (6 out of 71 children with suspected DLD at 2.5/3 years). The positive predictive value (PPV) was 40%, indicating that there was 40% risk to fail the L-screening after a positive CB-screening. As an overall measure of predictive validity, the diagnostic odds ratio was 8.57.

Table 6. Results of the canonical babbling screening (CB-screening) at 10 months and the language screening (L-screening) at 2.5/3 years

| <b>CB-screening</b>              | <b>L-screening</b> |                     | <i>Row sum</i>    |
|----------------------------------|--------------------|---------------------|-------------------|
|                                  | sDLD (positive)    | Not sDLD (negative) |                   |
| <b>Delayed CB (positive)</b>     | 6                  | 9                   | <i>15</i>         |
| <b>CB established (negative)</b> | 65                 | 836                 | <i>901</i>        |
| <i>Column sum</i>                | <i>71</i>          | <i>845</i>          | <i>Total: 916</i> |

The results of the L screening for the 93 children who participated only by sharing results on the regular L screening, showed that the prevalence of children with failed L screening result was comparable between the group who went through the CB screening (7.8%) and the group that did not participate in the CB screening (9%). This supports the generalizability of the results.

#### 5.4 STUDY IV

Regarding babbling assessment at 12 months, 12 (80%) out of 15 children were still not in the CB stage and half of the group (8 children, 53%) neither had dental/alveolar stops nor any true consonants. Mean number of different true consonants was 0.8 (range 0-4). For assessment of consonant production, five children were excluded due to low word production, <50% of the target words in the test. The remaining nine children had established 8.7 out of 10 target consonants and PCC-A was 89% (SD 0.17). For parent-reported expressive vocabulary at 18 months, mean number of expressive words was 20.4 (range 0-125) and six out of 15 (40%) children had results below the 10<sup>th</sup> percentile (7 words) according to the age-based Swedish norms. At 36 months, the mean number of expressive words was 408 (range 0-660) and three (20%) of the children had results below the 10<sup>th</sup> percentile according to the age norms for 28-month-old children. Mean composite scores for cognitive abilities was 100.4 (SD 14.1), for



language abilities 92.3 (SD18.9) and 91.5 (SD 11.7) for motor abilities. There were 3 (21%) out of 14 children who had scores below the normal range, 1 child (7%) in one subdomain and 2 (14%) children in two or three subdomains. There were no significant differences on any of the assessments between the group of children who lacked CB at 10 months and the group of children who had speech and language difficulties at age 3 years, identified by the language screening at 2.5/3 years.

Three children in the positive screened group had results that stood out from the rest. According to the informal comments given by parents, they expressed themselves mainly non-verbally or by acting out what they wanted (that is, pointing and showing) at both 18 and 36 months. The children had followed the regular program at the CHS during the first year. At least two of the children were referred to a SLP at 2 years of age and at age 3 years, the three children were either waiting for a developmental examination or had already undergone one during the preceding six months.

## **5.5 STUDY V**

Fifty consecutive child utterances and the parent's contingent responses for each of the 22 parent-child dyads were annotated and categorized from video recordings of free play situations between 10-month-old children and one of their parents. There was no significant difference in proportion of contingent responses after CB utterances and after pre-canonical vocalizations, neither when utterances from all children were summed up nor by dividing the utterances after each child's babbling stage (having reached CB stage or not). For contingent response types, there was no significant difference when all utterances were summed up. However, when divided by groups according to babbling stage, children who had not reached the CB stage (late babbling) received a higher proportion acknowledgment following a CB utterance ( $Z= 1.992$ ,  $p= 0.046$ ) than following a pre-canonical vocalization and higher proportion comments after pre-canonical utterances ( $Z= -2.207$ ,  $p= 0.027$ ). In the group of children who had reached the CB babbling stage, (typical babbling) there was a tendency for a higher proportion imitations/expansions after CB utterances than after pre-canonical vocalizations ( $Z= -1.648$ ,  $p= 0.099$ ).



## **6 DISCUSSION**

The present thesis project aimed to examine if an early CB screening procedure within the CHS would be useful to find children at risk for DLD, with the purpose of offer early intervention to children who would benefit from it. A prerequisite was reliable assessment methods for an easy administered screening procedure as well as follow-up assessment by a SLP. These aims will be discussed with consideration to the project starting points, the CB screening in the Swedish context, early detection, expressive language development and parent responsiveness. Points of perspective will then follow.

### **6.1 STARTING POINTS**

The present project builds on the foundations of the telephone project by Oller and colleagues from the 1990s (1998; 1999). It takes advantage of simple but still valid questions to parents about their child's babbling and it uses the age described as the cutoff age for considering a delay in babbling development. The proposed CB screening expands on the telephone project in a couple of ways (Oller, Eilers, et al., 1998; Oller et al., 2001; Oller et al., 1999). Instead of posing questions over the phone, the CB screening was performed within the regular CHS framework, and the questions about babbling were asked during an established visit at 10 months already including a developmental surveillance. The questions were asked by the CHS nurse who already knew the child and family. The assessment used for prediction is the language screening at 2.5/3 years already in use within the CHS (Miniscalco Mattsson et al., 2001; Westerlund & Sundelin, 2000). It also expands on the assessment for validation of the positive screened children by including consonant production alongside presence of CB and the follow-up assessments of the positively screened children are extended to age 36 months including consonant production, expressive vocabulary as well as assessment of general development.

The "CB screening" [Jollerboll] consisted of three validated CB questions to the parent on the child's babbling and an assessment by a SLP for the positively screened children. As mentioned above, the screening questions, formulated as open-ended questions to start parent's descriptions of the babbling with their own examples and words, were validated by Oller et al. (1999; 2001). On the other hand, the observation method that would fit the CB assessment by SLP was not yet validated. To examine the validity and utility of the observation method, the thesis project included two method studies for this basic purpose. An observation form was originally created within an international project of children with cleft palate, the Scandleft project (Lohmander et al., 2011). It has been used in different versions throughout the years

and is still under evaluation and adjusted after feedback from users and researchers. In the data collection of the present project, the version including a consonant chart for notation of consonants heard at least twice was used. The notations were then used for information of number of consonant inventory (different consonants and different true consonants). In a later version of the form, remembered syllables (including consonants) were noted freely out of memory right after the observation. The consonant inventory, and similar variables are thus measured from both methods. In Study I in this project, the comparisons between phonetic transcriptions of utterances from the recordings (Lohmander et al., 2011) and data from the observations forms of the same recordings were found to be comparable, while the latter was faster and simpler. However, the correlation of consonant inventory was low. Transcriptions may overestimate consonant inventory (Ramsdell et al., 2012) while noting syllables, on the other hand, have been reported to underestimate number of consonants (Willadsen et al., 2020).

## **6.2 THE “CB SCREENING” IN THE SWEDISH CONTEXT**

As the “CB screening” only identified a few of the children who failed the language screening at 2.5/3 years (low sensitivity), the usefulness as a universal screening within the CHS is not evident. The WHO recommendations for screenings can assist in balancing the evaluation of the CB screening (Wilson & Jungner, 1968). Two of the 10 recommendations, are two principles that regard the screening instrument itself. The first is the instrument’s acceptance to the population and the second the use of a suitable instrument, meaning a screening with satisfying validity. Since the “CB screening” is simple and easy to understand for both parents and health professionals, it would presumably be accepted by the CHS and the families taking part in it. However, the acceptance was not examined in the project. Regarding screening validity, there are a few aspects to consider. Primarily, the low sensitivity is problematic. It implies that there are many children who will pass the CB screening but still fail the language screening two years later (false negatives). The CB screening address children at a very young age, and sensitivity seems to improve with age at screening, whereas specificity remains high (Sansavini et al., 2021). The time interval between CB screening and the language screening used as golden standard, is also a factor. In general, it seems easier to identify children without language disorders than identifying those with the disorder, as many studies report higher specificity than sensitivity (Sansavini et al., 2021). Despite these examples of contributing factors, the low sensitivity is not satisfactory. Instead, one could consider the possibility of integrating the three CB questions [Jollerboll] in the already existing developmental surveillance at 10 months. Without adding time to the visit for parents or for the CHS nurse, the gain in information would be maintained. With a positive result on the questions, the CHS

nurse's decision-making on risk for DLD for individual children would be facilitated. In these cases, there is higher certainty that the child will develop DLD than without the CB questions, considering the PPV of 40% of the CB screening in the project compared to the prevalence of 7.8% of failing language screening. This will enable the CHS nurse to direct support to the children in need for it, which is in accordance with the aims and guidelines of the CHS (The National Child Health Care program, 2019). For the individual child (true positives), SLP assessment and early individualized intervention would be possible. The importance of follow-ups after screening was highlighted in a recent qualitative study on caregiver's experiences of developmental screening (Andersson, Miniscalco, & Gillberg, 2014). Parents requested conversations with a professional about the screening results, the next stage in development, and their role in facilitating progress (Traube et al., 2021). This could be taken care of in the SLP follow-up assessment performed within 2-3 months. The false positive screened children with a continued typical development would probably be dismissed after the SLP follow-up and thereby not afflicted with distress or negative experiences. However, these are presumptions and were not examined in the thesis. With the possibility of later identification at the 18-month- surveillance or 2.5/3 years screening, the false negative children would still be identified and the last WHO recommendation, that "Case-finding should be a continuing process and not a "once and for all" project" would be met (Wilson & Jungner, 1968, pp. 28-29).

### **6.3 EARLY DETECTION WITHIN THE CHILD HEALTH SERVICES**

The national program of CHS is regulated by the National Board of Health and Welfare, updated in 2014 (National Board of Health and Welfare, 2014). There are also web-based guidelines and regional supplements. The "CB screening" was performed within the CHS framework and was preceded by several steps of information and practice in recognizing CB during the first year of life, in the form of large-scale lectures, repeated and detailed information on babbling and the screening procedure and continuous contact by phone and email. The 10-month-surveillance in use, includes questions to parents on understanding of sentences and following instructions, there is no standardized material to be used according to the web-based guidelines. If the use of the CB questions would be tried at the 10-month-visit in a larger scale, added information on the babbling would be added and risk factors would be evident. Efforts of education and training to the CHS professionals would be needed. This would imply a cost, but since no equipment or material is needed the cost would be relatively small. Although not examined in a systematic way, the informal comments by the nurses during the project period, was that the screening procedure itself was easy to carry out and helped them to pay attention

to vocalizations and CB. Experience from a project where a checklist for communication and language was used at the 8-month-visit (Fäldt et al., 2021), conclude that the CHS nurses appreciated to have a material and a structure for the surveillance during the project. Deeper understanding of the CHS nurse perspective on asking the CB questions would be desirable.

#### **6.4 BABBLING AND EARLY EXPRESSIVE VOCABULARY**

The CB variables used in this project, CB onset before 10 months, presence of oral stops, anterior stops and specifically dental/alveolar stops, and number of different true consonants at 12 and 18 months reflect the current knowledge base on predictive CB (Lohmander et al., 2021; McDaniel et al., 2020; Willadsen et al., 2020). Despite the knowledge of these predictive variables in children with TD and in clinical groups (McCune & Vihman, 2001; Stoel-Gammon, 2011), the natural course of development of expressive language for children with delayed CB without known medical condition has not yet been studied.

A first reflection is that delayed CB is a rare condition. The prevalence in the present study of children without medical diagnosis was 1.4% which was lower than the estimation of 3.1% in the telephone-project by Oller and colleagues (Oller et al., 1998; 1999), which was based on a high-risk population. Including children with known clinical diagnosis the prevalence was 6% in that study, which was expected for this theses project as well, where, for example, hearing loss, cleft palate, or neurodevelopmental delays were not an exclusion criterion. Despite this, there were no children from any of these risk groups in the study. This probably lowered the prevalence to some extent.

The children identified as having delayed CB at 10 months (failed CB screening) did not use the predictive CB variables at 12 months, indicating a risk for expressive language delays. At 18 months, the risk indication was still present according to lack of some CB variables. Dental/alveolar stops had emerged for most of the children, but the mean number of true consonants was still lower than described age references (Jones, Chapman, & Hardin-Jones, 2003; Lohmander et al., 2011; Scherer, Williams, & Proctor-Williams, 2008). The development of number of different true consonants from 12 to 18 months (0.8 and 4.5 respectively) was similar to the development for children in clinical groups, for example children with hearing loss (Persson, Flynn, Miniscalco, & Lohmander, 2021) and cleft palate (Lohmander et al., 2021). True consonants include consonants that cannot stand on their own to form a syllable, in contrast to for example semivowels which are not included. In that sense, it is natural to use this measure in detecting nuances in the consonant production in babbling.

Glottals, which are not included in true consonants either, showed low agreement between observation and transcription in study I in the CB project, suggesting that these consonants are hard to observe in a reliable way. Glottals and posterior consonants are not correlated to expressive vocalizations and articulation measures, hence not predictive and thereby not included in the reasoning here.

Mean number of expressive words at 18 months in the present project, as measured by the Swedish version of the parent questionnaire McArthur-Bates Communicative Developmental Inventory (MCDI) (Fenson et al., 1993), was below the 10<sup>th</sup> percentile for about half of the children compared to the age-based norms and thus, considered as delayed (Berglund & Eriksson, 2000). For 80% of the children the result was below the 50<sup>th</sup> percentile. At 36 months, we could not establish reliable comparisons since the age-based norms only reaches up to 28 months. To have some point of reference, the norm for 28 months was used for the 36-month-old children. Interestingly, the comparison showed similar group means, indicating that the CB delayed children, had equal mean number of words as children six months younger. A group of typically developing children in the same population would have given an age-reliable reference and presumably there would have been a difference in mean number of words. One also must consider that the expressive vocabulary was reported by parents. A bias of overestimation of the vocabulary has been discussed, more specifically from parents with lower level of education (Law & Roy, 2008). This bias would probably not affect the present project, for most of the participating children, at least one of the parents had education above (Swedish) high school.

For 3 children, 20% of the positively screened children, the delayed CB at 10 months was an early sign of a general developmental disability which could be noticed consequently in both assessments and the informal comments by parents. The limited use of protophones during the first living year, including the most mature type of CB utterances, made these three children miss out on the described preparation phase for speech and language use, described as a basis for continued development (Long et al., 2020; Oller et al., 2021).

## **6.5 PARENT RESPONSIVENESS**

In the present project, we have taken advantage of the parents' recognition of CB onset in the screening process and of the responsive approach in the planning of intervention for children with delayed CB.

Some may say that early identification is a first step towards the development of efficient interventions. Others would say that screening is not justified if there is no efficient intervention. The opinion to wait and see what happens with a child with early signs of speech or language difficulties has been described as outdated (Capone Singleton, 2018) Instead, a proactive attitude with specialist referral and intervention has been recommended. The result from this project supports this approach. With the possibility of early identification, already after the CB screening at 10 months, parental guidance and intervention would be brought forward. Although there is no existing intervention method for toddlers with suspected language problems, both well-established favorable strategies that parents to TD children use and evidence-based methods for older children with disorders can be starting points. According to a report on evidence-based practice in the habilitation services in Sweden (Eberhart et al., 2017), there is strong evidence for an association between parents' responsive approach in communication and both current and future language abilities in the child. Guiding parents towards a responsive approach can affect parental responsiveness and enhance language development for the child (Eberhart et al., 2017). With facilitating word acquisition as a goal, setting off the social feedback loop between parent and child would be desirable (Gros-Louis & Miller, 2018). For children with TD, parents respond to more mature utterances, for example a CB utterance, with a language-stimulating utterance which in turn, encourages the child to make another mature utterance. As the results indicate, in clinical groups with children who use only few mature CB utterances, parents do not use the language stimulating response types fully. Thereby, neither child nor parent succeed in setting off the feedback loop. In intervention, parents could be guided to pay attention to the child's utterance types as well as different ways to respond to them. Aspects of child directed speech that are associated with word learning would be suitable goals, for example pause duration in turn-taking (Marklund, Marklund, Lacerda, & Schwarz, 2015) and enhanced vocal hyper-articulation (Marklund, Marklund, & Gustavsson, 2021). Paying attention to non-verbal communication occurring alongside the utterances and responding to them might be another goal.



## 7 CONCLUSIONS

Observation, and making notes on a systematic observation form, is a valid method to assess presence of CB and consonant variables such as oral stops, anterior stops, and dental/alveolar stops. Presence of these variables have been pointed out as predictive of later expressive language development and are therefore interesting targets for a babbling observation.

The CB screening had high specificity, indicating that most children without risk for DLD at 2.5/3 years were dismissed already at 10 months. However, the sensitivity was low and not satisfactory for a useful universal screening. Continued delay at 18 months was found in less than half of the children with positive screening result and for about one fifth the delay persisted at 36 months. For these at-risk children the delay was to be part of a more general developmental disability. To take advantage of the systematic information gained by the CB questions to parents, inclusion of these valid questions in the 10-month-surveillance was recommended. Quick follow-up assessment by a specialist after identification at 10 months, an “intervention package” with information on the result, the next stage in development and how to reach it, would be of value for these families.

Parents to children in a clinical group at 10 months, used the same proportion of contingent responses after vocalization utterances and the more advanced CB utterances, which differs from reports on parents to children in non-clinical groups. A responsive approach does not seem to be used to its full potential by parents of children with delayed CB at 10 months.



## 8 POINTS OF PERSPECTIVE

### 8.1 CLINICAL IMPLICATIONS

There are different possible ways to implement the results from this project, including the CHS, speech and language pathology (SLP) and the collaboration between them. For the CHS, it could imply adjusting information in the national guidelines, consideration of implementing the CB questions at 10 months and at 6 months adjust the current question on nuanced babbling (figure 1). Learning and training activities would be needed to prepare for implementation as well as closer cooperation between CHS and SLP for children with delayed CB. As suggested in figure 1, the arrangements for children who do not use CB could include discussion within the CHS team to pinpoint the child’s general development and to survey the special needs of the child and family. Support and guidance to the family at this stage could take advantage of the newly developed language-enhancing materials for use within the CHS in many regions. For the SLP clinics, implication would mean adjusting intervention programs, preferably with responsiveness as goals, to this age-group with no earlier known medical conditions. A referral to SLP for an “intervention package” at around 11 to 12 months, including in-depth CB assessment as well as individualized guidance on responsive approach, would be a way to offer early contact with a specialist. It would be beneficial to include a follow-up visit by the SLP for further guidance and assessment and cooperation with the CHS at 18 months.

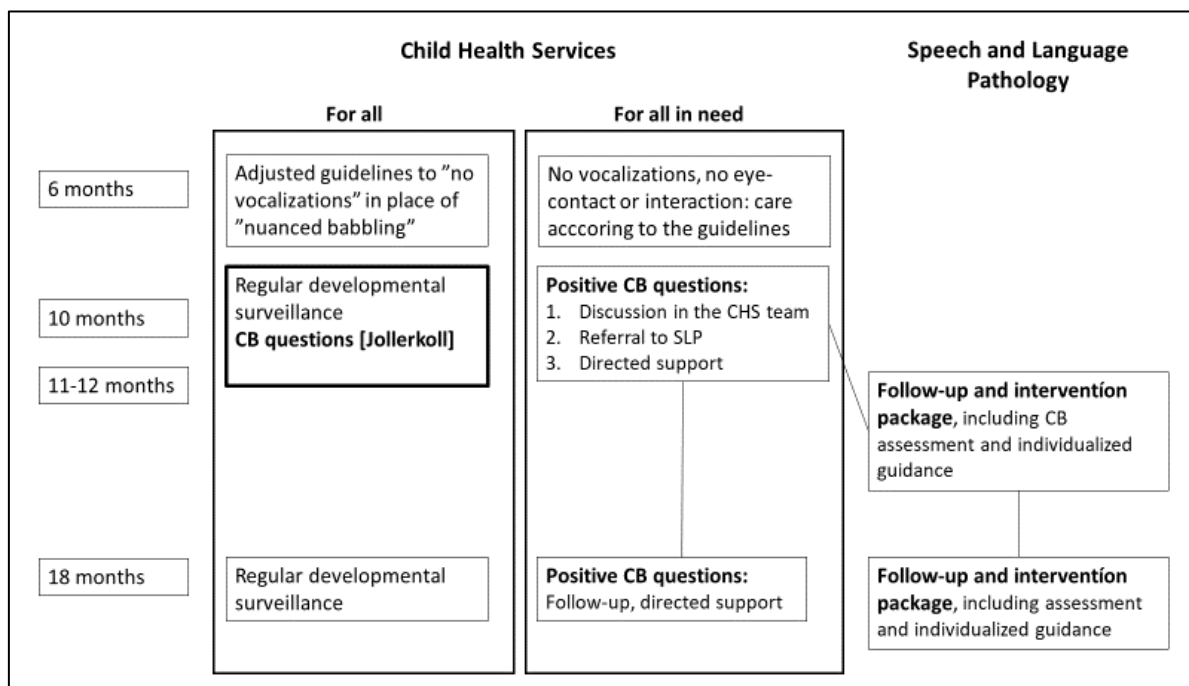


Figure 1. Suggestion for implication on the thesis results within Central Health Services (CHS) and Speech and Language Pathology (SLP)

## **8.2 FUTURE RESEARCH**

Deeper understanding of the trajectories for positively screened children could be gained in a continued follow-up at school entry.

An evaluation of the CHS surveillance of early babbling for children with and without known risk factors would be of interest, considering that communication and language is included in the regular developmental surveillance visits during the first living year.

To realize the planned intervention study that was not carried out in this thesis project would bring additional information. A comparison of intervention after early detection at 10 months with intervention at a later stage would increase knowledge of the value of early identification as well. In general, it would be interesting to investigate parents' responsive approach and the social feed-back loop for children in clinical groups at different stages in language development.

To include babbling in different situations, for example vocal play when the child is on its own and in interaction, with a parent responding verbally or only non-verbally, in studies of early expressive language in children in clinical groups, would contribute with information needed for planning individualized parent-directed intervention.

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