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BABBLING, SPEECH AND LANGUAGE IN CHILDREN WITH NEUROLOGICAL DISABILITIES – DEVELOPMENT, VALIDITY OF MEASURES AND EFFECT OF INTERVENTION

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Babbling, Speech and Language in Children with Neurological Disabilities – Development, Validity of Measures and Effect of Intervention
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POPULAR SCIENCE SUMMARY OF THE THESIS

Da-DA-da! Dededada. Eh! Bab. Di. Ba-ba. Dat! Hada! Babadedi! Mmmmm.

When an infant starts babbling it is an important milestone, often noted by parents and others close to the child. Parents pay especially close attention to babbling consisting of syllables of consonants and vowels (such as ma-ma or da-da). What they may not know is that there are associations between how children babble and their later speech and language development. These associations have been used by researchers trying to identify children who may develop speech and language difficulties during the preschool years. Speech and language difficulties in children can challenge, for example, their learning of new words, or their articulation of words, leading others to not understand them. Sometimes, the problems lead to life-long difficulties in making themselves understood. Some children never develop spoken language. This is especially common in children who have neurological disabilities, for example cerebral palsy or Down syndrome.

Although many children with neurological disabilities have difficulties communicating, we do not know exactly how speech and language skills develop in this group of children. There is also limited knowledge on how to best treat the difficulties. Examining infant babbling could be a method for predicting speech and language disorder in this group. One challenge is, however, that the measures used would have to be valid, which is something that has not been thoroughly examined. The aim of this thesis was to evaluate the validity of a new and simpler measure of babbling and to examine babbling, speech and language over time in a group of children with neurological disabilities. The thesis also contains a treatment study, examining the effect of a language training method for children with cerebral palsy.

The results showed that the new, simplified babbling measure CBR^{UTTER} gave similar results as other established babbling measures, indicating that it can identify children whose babbling is delayed. The children with neurological disabilities examined in this thesis had delayed babbling to a higher extent than children without disabilities. At ages five and seven, most of the children with neurological disabilities had some kind of speech or language disorder. Their parents reported that they had big difficulties communicating in their daily life, but the number of treatment sessions that children had received during the last year was often low. In the treatment study, parents of four children with cerebral palsy and speech difficulties learned a vocabulary training technique, which they used at home daily for five weeks. The training results were good for two of the children, indicating that it may be effective for children with cerebral palsy.

For many families, children's language development is a source of joy and amazement, but this is not always the case when the child has a neurological disability. This thesis show that it is important to identify speech and language disorders in children with disabilities and that treatment may reduce the difficulties.

POPULÄRVETENSKAPLIG SAMMANFATTNING

Da-DA-da! Dededädä. Äh! Dädä di-di. Däd. Di. Bä-bä. Dät! Hadä! Bäbädidi. Mmmm.

Barns första joller är en viktig milstolpe som ofta uppmärksammas av föräldrar och andra i barnets omgivning. Särskilt noga lägger omgivningen märke till stavelsejoller, det vill säga joller som består av konsonanter och vokaler (t ex ma-ma eller dädädä). Mindre känt är kanske att det finns samband mellan hur barn jollrar och den senare tal- och språkutvecklingen. Forskare har försökt använda de här sambanden för att förutsäga vilka bebisar som kommer att utveckla talsvårigheter i förskoleåldern. Talsvårigheter hos förskolebarn kan till exempel innebära svårigheter att hitta ord eller uttalssvårigheter som gör att omgivningen får svårt att förstå vad barnet säger. Ibland är svårigheterna så stora att barnet under hela livet har svårigheter att uttrycka sig. Vissa barn utvecklar aldrig ett talat språk. Det här är ett särskilt stort problem hos barn som har neurologiska funktionsnedsättningar, som cerebral pares eller Downs syndrom.

Trots att många barn med neurologiska funktionsnedsättningar alltså har svårt att kommunicera, vet vi idag inte exakt hur tal- och språkförmågan utvecklas hos dessa barn. Dessutom är kunskapen begränsad om hur svårigheterna bäst ska behandlas. Att undersöka hur barnen jollrar skulle kunna vara en metod för att förutsäga talsvårigheter hos den här gruppen. Utmaningen är att måtten som används för att undersöka joller måste vara träffsäkra, vilket inte har undersökts ordentligt tidigare. Syftet med den här avhandlingen var att utvärdera träffsäkerheten hos ett nytt och enklare sätt att mäta stavelsejoller och att undersöka joller, tal och språkförmåga över tid hos en grupp barn med neurologiska funktionsnedsättningar. Avhandlingen innehåller också en behandlingsstudie, som undersöker effekten av en träningsmetod för små barn med cerebral pares och talsvårigheter.

Resultaten visade att det nya, förenklade jollermåttet CBR^{UTTER} gav liknande resultat som andra etablerade jollermått, och alltså träffsäkert kan hitta barn som saknar stavelsejoller. Barnen med neurologiska funktionsnedsättningar hade i högre uträckning försenad utveckling av joller än barn utan funktionsnedsättningar. Vid fem och sju års ålder hade de flesta av dem någon form av tal- eller språkstörning. Föräldrarna rapporterade att barnen hade stora svårigheter att kommunicera i vardagen, men antalet behandlingstillfällen som barnen hade genomgått under det senaste året var ofta få. I behandlingsstudien fick föräldrar till fyra barn med cerebral pares och talsvårigheter lära sig en teknik för att träna barnets ordförråd. Träningen, som genomfördes av föräldrarna under fem veckor, gav goda resultat för två av barnen. Den här typen av träning kan alltså fungera för barn med cerebral pares, men passar förmodligen inte alla.

För många familjer är barns språkutveckling en källa till glädje och förundran, men det är inte självklart när barnet har en neurologisk funktionsnedsättning. Den här avhandlingen visar att det är viktigt att uppmärksamma och identifiera tal- och språksvårigheter hos barn med funktionsnedsättningar och att det kan vara värt att sätta in extra behandling för att mildra problemen.

ABSTRACT

Children with neurological disabilities (ND) such as cerebral palsy, Down syndrome or other genetic syndromes often have speech and language disorder. In Sweden, children with ND often receive team-based habilitation services – with teams including speech and language pathologists – from an early age, but even so, many things are still unknown when it comes to the expected development of speech and language abilities. Furthermore, service delivery for speech and language difficulties has rarely been studied in the Swedish context and even internationally there is a lack of studies examining the effects of intervention.

Babbling is an important precursor to speech and has been studied in many groups of children at risk of speech and language disorder. Thus, it shows promise as a field of study for children with neurological disabilies as well. One commonly studied babbling milestone is the onset of canonical babbling, when children produce speech-like syllables consisting of consonants and vowels. However, the most frequently used canonical babbling measure – the canonical babbling ratio (CBR) – has not been thoroughly validated. This thesis aimed to validate the CBR measure, to examine babbling, speech and language in a group of children with ND and to evaluate an intervention for young children with ND, specifically cerebral palsy.

The thesis consists of five studies. In study I and II, a new and simplified version of the CBR measure (CBR^{UTTER}) was validated. It was found to be valid compared to other versions of the CBR and to babbling observation, suggesting it as an alternative to more laborious measures. Study II also evaluated the CBR criterion for the canonical babbling stage. A child is commonly considered to be in the canonical babbling stage when 15% or more of their babbling is canonical, that is a CBR of ≥ 0.15. Study II suggests that 0.14 may be a more appropriate criterion and emphasizes the need for proper discussion among babbling researchers on criteria for having entered the canonical babbling stage.

In study I, III and IV, babbling, speech and language were examined at ages 1, 5 and 7 in a group of 18 children with neurological disabilities who received habilitation services from an early age. Compared to data from typically developing children, babbling milestones were delayed in the group of children with ND. At 5 and 7 years of age, speech and language disorder was very common, with only one participant presenting with results at age level on all measures. The severity of speech and language disorder was very varied, but a majority of participants had severe communication activity limitations. Despite this, parents rated the frequency of received SLP services as low. The results accentuate the need to closely follow speech and language development in children with ND, and to provide intervention as needed.

In study V, a parent-implemented intervention for speech/language disorder was examined in young children with cerebral palsy. In a single case A-B study, four children received an intervention aimed at improving expressive vocabulary using a focused stimulation technique. Two children clearly improved their expressive vocabulary following the intervention, one showed less clear gains and one did not improve. Focused stimulation may thus be successful

in children with cerebral palsy and speech/language difficulties and could be a valuable addition when it comes to SLP intervention options in children with neurological disabilities.

In summary, this thesis presents new insights on the validity of the CBR measure, emphasizes the need to assess speech and language in children with neurological disability, and suggests an intervention for young children with cerebral palsy.

SAMMANFATTNING

Barn med neurologiska funktionsnedsättningar (neurological disabilities, ND) som cerebral pares och genetiska syndrom (t ex Downs syndrom) har ofta tal- och språkstörning. Barn med neurologiska funktionsnedsättningar får ofta insatser från tvärprofessionalla team inom habiliteringen, bland annat av logopeder, men hur utvecklingen av tal- och språkförmågor ser ut över tid är fortfarande i hög utsträckning okänt. Det har inte heller tidigare undersökts hur logopedinsatser utformas för barn med ND i Sverige och överlag finns det förhållandevis få studier som undersöker effekten av intervention för barn med ND och tal/språkstörning.

Joller är en viktig föregångare till tal och har därför studerats i många olika grupper av barn som riskerar tal- och språkstörning. Det skulle alltså kunna vara ett lovande område att studera även hos barn med neurologiska funktionsnedsättningar. En viktig milstolpe i jollerutvecklingen är debuten av kanoniskt joller, där barn producerar talliknande stavelser bestående av konsonanter och vokaler. I klinisk jollerforskning används ofta måttet ”andel kanoniskt joller” (canonical babbling ratio, CBR), men detta mått har inte validerats efter att det introducerades på 90-talet. Syftet med den här avhandlingen var att validera CBR-måttet, att undersöka joller, tal och språk hos en grupp barn med neurologiska funktionsnedsättningar och att utvärdera en intervention för små barn med en typ av ND, nämligen cerebral pares.

Avhandlingen består av fem delarbeten. I studie I och II validerades en ny och förenklad version av CBR-måttet (CBR^{UTTER}). Det nya måttet visade sig vara valitt jämfört med andra versioner av CBR och jämfört med bedömningar baserade på observation. CBR^{UTTER} kan alltså vara ett alternativ till andra, mer tidskrävande CBR-mått. Studie II utvärderade också gränsen för när ett barn anses vara i det kanoniska jollerstadiet. Vanligtvis används gränsen ≥ 0.15, det vill säga ett barn anses vara i det kanoniska jollerstadiet när 15% eller mer av jollret är kanoniskt. Resultaten i studie II tyder på att 0.14 kan vara en bättre gräns, och understryker behovet av diskussion bland forskare om vilka kriterier som ska användas för att avgöra om ett barn är i det kanoniska jollerstadiet eller inte.

I studie I, III och IV undersöktes joller, tal och språk vid 1, 5 och 7 års ålder hos en grupp barn med neurologiska funktionsnedsättningar som tidigt fick insatser från habiliteringen. Jämfört med data från typiskt utvecklade barn var utvecklingen av joller försenad hos barnen med ND. Vid fem och sju års ålder var tal- och språkstörning väldigt vanligt – endast en deltagare hade åldersadekvata resultat på samtliga test. Tal- och språksvårigheterna var av varierad svårighetsgrad, men en majoritet av deltagarna hade stora kommunikativa aktivitets-begränsningar. Trots detta rapporterade många föräldrar att barnen sällan fick insatser från logoped. Resultaten belyser vikten av att noga följa tal- och språkutvecklingen hos barn med neurologiska funktionsnedsättningar och ge insatser vid behov.

I studie V undersöktes en föräldraimplementerad intervention för små barn med cerebral pares och tal/språkstörning. I kontrollerade fallstudier (s.k. A-B design), genomgick fyra barn en intervention med syfte att öka det expressiva ordförrådet med hjälp metoden fokuserad språkstimulering. Två deltagare förbättrades tydligt under interventionen, en förbättrades i viss

mån och en uppvisade inga förbättringar. Fokuserad språkstimulering kan således vara effektiv för barn med cerebral pares och tal/språksvårigheter och utgör ett möjligt verktyg för logopedbehandling av barn med neurologiska funktionsnedsättningar.

Sammanfattningsvis erbjuder den här avhandlingen nya insikter om validiteten för måttet CBR, lyfter fram behovet av att bedöma tal och språk hos barn med neurologiska funktionsnedsättningar samt föreslår en intervention för små barn med tal/språkstörning associerad med cerebral pares.

LIST OF SCIENTIFIC PAPERS

I. Nyman, A., & Lohmander, A. (2018). Babbling in children with neurodevelopmental disability and validity of a simplified way of measuring canonical babbling ratio. *Clinical Linguistics & Phonetics, 32*(2), 114-127.

II. Nyman, A., Strömbergsson, S., & Lohmander, A. (2021). Canonical babbling ratio – Concurrent and predictive evaluation of the 0.15 criterion. *Journal of Communication Disorders, 94*, 106164.

III. Nyman, A., Strömbergsson, S., Lindström, K., Lohmander, A., & Miniscalco, C. (2021). Speech and Language in 5-year-olds with Different Neurological Disabilities and the Association between Early and Later Consonant Production. *Developmental Neurorehabilitation, 24*(6), 408-417.

IV. Nyman, A., Miniscalco, C., Lindström, K., & Strömbergsson, S. Speech and language development and received services between 5 and 7 years of age in children with early diagnosed neurological disabilities. *Manuscript.*

V. Nyman, A., Miniscalco. C., Lohmander, A., & Strömbergsson, S. Expressive vocabulary intervention for four 2–3-year-old children with cerebral palsy and speech/language difficulties: a single case A-B study. *Manuscript*

CONTENTS

LIST OF ABBREVIATIONS

AAC	Augmentative and alternative communication
AUC	Area under the curve
CAS	Childhood apraxia of speech
CB	Canonical babbling
CBR	Canonical babbling ratio (see table 3, p 26 for definitions of different versions of the CBR)
CI	Confidence interval
CP	Cerebral palsy
DS	Down syndrome
FXS	Fragile X syndrome
LR+	Positive likelihood ratio
ND	Neurological disabilities
PCC	Percentage consonants correct
PIMD	Profound intellectual and multiple disabilities
PWS	Prader-Willi syndrome
ROC curve	Receiver-operating characteristics curve
SD	Standard deviation
SECDI-w&g	Swedish Early Communicative Development Inventory – words & gestures
SECDI-w&s	Swedish Early Communicative Development Inventory – words & sentences
SLP	Speech-language pathologist
SSD	Speech sound disorder
SVANTE	Swedish Articulation and Nasality Test
T1	Time point 1
T2	Time point 2
T3	Time point 3
TOM	Therapy Outcome Measures

1 INTRODUCTION

I came into this PhD journey with eight years of clinical experience as a speech-language pathologist (SLP) within the habilitation services, a multi-professional service provided for people with disabilities in Sweden. My clinical work had mainly concerned children with movement disorders and intellectual disability. One motivation behind my decision to become a doctoral student was that I wanted to learn more about how to best support these groups as an SLP. I knew from clinical experience that these children were often referred to the habilitation services early in life, some already diagnosed with for example cerebral palsy, Down syndrome or other genetic syndromes, others with no known medical cause for their often severe developmental delays. As the children grew older, the proportion with a known medical cause of their disability would increase and many children would later end up with diagnoses such as intellectual disability or autism spectrum disorder in addition to the medical diagnoses. In this thesis, I use the term "children with neurological disabilities" for this patient group. I chose this instead of the more common "neurodevelopmental disability". Although diagnoses such as cerebral palsy and Down syndrome are sometimes included under the umbrella of "neurodevelopmental disability", it is most often used for diagnoses such as autism, ADHD and developmental language disorder. As these conditions are not primarily in focus for the thesis, I opted for another term, that could reflect the diversity of my group of interest – children receiving habilitation services from infancy.

When entering doctoral education, I became a part of a research group focusing on the early development of speech and language in children with different disabilities or risk factors. One major interest of the research group was babbling as a precursor to speech. As earlier research has shown, babbling is in many ways associated to the later speech and language development and infant vocalizations have therefore been examined quite thoroughly in for example children with cleft palate and hearing loss. As I will show, examinations of babbling in children with ND are however less common (even more so at the time when this project started).

This PhD project and thesis is thus a result of two converging fields: speech and language in children with neurological disability and babbling as a precursor to speech in clinical risk groups. Although I started out with an attempt to investigate babbling in neurological disabilities in particular, I later developed an interest in the methodology of infant vocalization research. Thus, this thesis ended up spanning quite a wide field – including characterization and treatment of speech and language disorders in children with neurological disabilities, babbling as a precursor to speech in children with neurological disabilities and validity of commonly used measures in clinical babbling research.

2 LITERATURE REVIEW

2.1 BABBLING AS A PRECURSOR TO SPEECH AND LANGUAGE

When babies start to vocalize and babble, it attracts the attention of parents and others in their vicinity. The sounds a baby makes have a way of engaging the listener, who often attributes meaning to the vocalizations. Despite this, babbling was for some quite some time during the 20th century viewed as unconnected to speech. When Vihman and colleagues (1985) presented data on the longitudinal development of babbling and speech in nine infants, it took the form of an argument against this common believed discontinuity hypothesis – based on the theories of Roman Jakobson. Contrary to the discontinuity hypothesis, Vihman and colleagues found that babbling and words developed together and that although the infants showed different preferences in the consonant sounds used, the same consonant sounds were used in both babbling and words. In the same year, Stoel-Gammon (1985) showed that the consonant sounds commonly produced by 34 children during their second year were the same consonant sounds as previously reported as common in babbling. Later research on typically developing children has further strengthened the importance of babbling as a precursor to speech. Associations have been found between early consonant use and later expressive vocabulary (McCune & Vihman, 2001; McGillion et al., 2017) as well as later phonological proficiency (Vihman & Greenlee, 1987).

Although there are different models for describing infant vocalizations during the first year of life, there is a consensus on the general course of development. Here, focus will be on the development as described by Oller and colleagues (e.g., Oller, 1980; Oller, 2000; Oller et al., 1999). Other models for the development of babbling has been suggested by for example Stark and colleagues (e.g., Stark, 1980) and Koopmans-van Beinum and colleagues (e.g. Koopmans-van Beinum & van der Stelt, 1986).

The model of Oller and colleagues focuses on so called protophones. Protophones are vocalizations produced by infants, which are not reflexive (e.g. hiccups, burps) or a fixed emotional signal (i.e. cries, distress sounds, laughter). Formerly believed to be relatively rare during the first months, recent research has revealed that infants in fact use protophones from a very early age (Oller et al., 2021). In the first months (referred to by Oller and colleagues as the Phonation stage), protophones come in the form of so called quasi-vowels, i.e., vowel-like sounds produced with the vocal tract at rest. Quasi-vowels are produced with normal phonation, but without articulation, thus resulting in a sound less well-formed than vowels. In the primitive articulation stage (at 1–4 months of age), the infant begins moving the articulators, often resulting in so called gooing – posterior velar-like sounds. At 3–8 months of age, infants enter the Expansion stage, producing full vowels as well as sqeals and raspberries. At this stage, infants also begin transferring from an open to a closed vocal tract, producing syllable-like sounds called marginal babbling. Marginal babbling consists of consonant-like and vowel-like sounds, but with a slow transition between them, and thus less speech-like. In contrast, canonical babbling (CB) consists of speech-like syllables with a rapid transition between

consonant and vowel, such as /ba/ or /dædædæ/. CB usually emerges at 6–8 months (Eilers et al., 1993; Morgan & Wren, 2018). CB can consist of one or many syllables and can be reduplicated (/baba/) or variegated (/baga/). In earlier research, reduplicated and variegated babbling were considered two separate stages, but later research has shown that they overlap considerably (Morgan & Wren, 2018; Smith et al., 1989).

Drawing upon research on the importance of babbling, researchers in speech-language pathology and related fields have examined early vocalizations in children with risk of developing speech and language disorders. Indeed, delayed or deviant development of babbling has been found in different types of childhood disabilities and risk factors, including hearing loss (Moeller et al., 2007a; Moeller et al., 2007b), cleft palate (Chapman et al., 2001; Lohmander et al., 2011; Scherer et al., 2008), and prematurity (D'Odorico et al., 2011; Strandberg et al., 2022; Törölä et al., 2012) as well as in children later diagnosed with autism (Patten et al., 2014; Roche et al., 2018), expressive language delay (Fasolo et al., 2008; Stoel-Gammon, 1989) and different speech sound disorders (Overby et al., 2020a; Overby et al., 2020b).

Two important aspects of babbling that have interested researchers are the development of syllable structure and the expansion of the consonant repertoire. Although there are considerable variation between infants, the development of the consonant repertoire seems to follow a general pattern, with plosives (especially the anterior t/d and p/b), nasals and glides as early emerging sounds and fricatives, liquids and affricates as later developing sounds (McCune & Vihman, 2001; Morgan & Wren, 2018; Stoel-Gammon, 1985). Researchers have been interested both in the number of different consonants used and in the use of specific types of consonants, for example plosives or dental/alveolar plosives[1] (see for example Klintö et al., 2014; Lohmander & Persson, 2008). The concept "true consonants" is sometimes used in research on early consonant production. Glottals and glides are not considered true consonants, due to their vocalic nature (McCune & Vihman, 2001). Another common adaption when tallying consonants in a young child's repertoire is to disregard voicing, that is, to consider voiced and voiceless realizations of the same articulation place and manner (for example /t/ and /d/) as one consonant. The rationale for this is that the voiced-voiceless distinction is not considered established at this young age (McCune & Vihman, 2001).

When it comes to the development of syllable structure, canonical babbling has been a major focus. As the onset of CB is both easily recognizable for parents and rarely delayed, it is considered a suitable variable for clinical babbling research (Oller et al., 1998). No difference has been found in CB onset between languages or between multilingual and monolingual infants (Lee et al., 2018; Oller et al., 1997). An onset later than 10 months of age is considered a clear sign of delay (Oller et al., 1999).

[1] That is, /t/ and /d/. In this thesis, the term "dental plosives" will be used, as /t/ and /d/ are produced with dental place of articulation in Swedish.

As in typically developing children, different babbling variables are related to later speech and language also in children with disabilities and other risk factors. Measures of consonant inventory in early speech has been shown to be related to vocabulary in children with hearing loss (Persson et al., 2021b) and children born prematurely (D'Odorico et al., 2011). For children with cleft palate, babbling consonant inventory measures are related to later consonant production (Chapman et al., 2003; Klintö et al., 2014; Lohmander & Persson, 2008), vocabulary (Chapman, 2004; Chapman et al., 2003) and sentence length (Chapman, 2004). Syllabic complexity have been shown to be related to the later vocabulary in children with expressive language delay (Fasolo et al., 2008), delayed CB onset (Oller et al., 1999) and premature birth (D'Odorico et al., 2011) as well as later consonant production in children with hearing loss (Moeller et al., 2007b) and cleft palate (Chapman et al., 2003). Finally, syllabic complexity has been found to be related to later results on language tests for children with expressive language delay (Whitehurst et al., 1991) and neonatal risk factors (Jensen et al., 1988).

Thus, there are associations between babbling and later speech and language not only in children with typical development, but also in children with clinical risk factors for speech and language disorder. These associations may, however, be hard to interpret, as babbling and infant vocalizations may be studied using many different methods.

2.2 METHODOLOGY IN CLINICAL RESEARCH ON INFANT VOCALIZATIONS

2.2.1 Methods

Phonetic transcription – using IPA symbols – has often been used for babbling analysis (e.g., Smith et al., 1989; Paul & Jennings, 1992 McCune & Vihman, 2001; D'Odorico et al., 2011; Fasolo et al., 2008; Lohmander et al., 2011; Moeller, Hoover, Putman, Arbataitis, Bohnenkamp, Peterson, Wood, et al., 2007; Scherer et al., 2008;). Based on transcription, both syllable shapes and consonant inventories may be analyzed. Transcription has, however, been critized as a babbling measure as especially non-canonical utterances are difficult to transcribe reliably (Ramsdell et al., 2007). As non-canonical vocalizations lack many of the basic traits of speech, for example rapid transitions between sounds, they are not easily represented with symbols design to capture human speech (i.e., the IPA alphabet). Furthermore, transcription over-estimates the number of sounds and combinations of sounds in an infant's repertoire, as compared to more naturalistic types of analysis (Ramsdell et al., 2012).

In contrast to phonetic transcription, categorization and coding procedures offer a possibility to analyze different types of infant vocalization without forcing an adult-like model upon them, which is especially useful when analyzing syllable shapes or emerging syllable shapes. The most well-known model for categorization is the infraphonological model, developed by Oller and colleagues (Oller, 2000; Oller et al., 1994). In infraphonological coding, vocalizations are classified as full vowels, quasi-vowels, canonical syllables and marginal syllables. Other categories of vocalizations may also be classified, for example raspberries, squeals and growls (Oller et al., 1994). Other examples of coding systems for babbling include coding for

phonation and number of articulatory movements (Schauwers et al., 2004, based on Koopmans-van Beinum & van der Stelt, 1986) and the Stark Assessment of Early Vocal Development (Nathani et al., 2006).

Coding of babbling variables may also be done in real time (Bass-Ringdahl, 2010; Belardi et al., 2017; Patten et al., 2014; Ramsdell et al., 2012; Willadsen et al., 2022; Willadsen et al., 2020). Sometimes referred to as naturalistic listening, this method includes categorizing utterances or syllables as canonical or non-canonical in real time while watching a recording of an infant, often using a software. If specific use of consonants (or consonant-vowel combinations) are analysed using naturalistic listening, they are often noted afterwards, based on the listeners overall impression. This has been shown to render inventories similar to parent report (Ramsdell et al., 2012).

Another method relying on overall impressions is home or laboratory observations. Here, a trained observer makes judgements as to whether certain babbling milestones are present in an infant's vocalizations, based on observation of the infant (on site or from recordings). Observation may be used as sole method (Lohmander et al., 2017a; Löfkvist et al., 2020) or together with parent report (Eilers & Oller, 1994; Eilers et al., 1993). Observation has been found to be a valid method for assessing consonant variables in babbling, compared to phonetic transcription (Lieberman & Lohmander, 2014). It has also been shown to be valid for assessing CB status, compared to counting in real time (Lohmander et al., 2017a).

Information on babbling may also be obtained from parental report. Oller and collagues (1998) used a series of open-ended and directed questions to examine canonical babbling during telephone interviews with parents. Parental reports on CB status were found to be accurate to a high extent, especially when parents responded to the open-ended questions. Lieberman and colleagues, using the same set of questions, found that 80% of infants whose parents reported no canonical babbling were confirmed as not having CB in an SLP observation (Lieberman et al., 2022). Another way of obtaining parental reports on babbling is using interview instruments such as the Vocal Development Landmarks Interview (Moeller et al., 2019).

2.2.2 Measures

In addition to different methods of babbling analysis, there is also a variety when it comes to the measures used to quanitify babbling. The Canonical Babbling Ratio (CBR) is a commonly used such measure. Developed by Oller and colleagues (Oller & Eilers, 1988; Oller et al., 1994), the CBR is a measure of the proportion of a child's vocal productions that is canonical. In the original version, later named CBR^{utt} by Molemans and colleagues (2012), the number of canonical syllables in a child's production is divided by the total number of utterances (Oller & Eilers, 1988). In an adaption, later referred to as CBR^{syl}, the number of canonical syllables is divided by the total number of syllables. Both CBR^{syl} and CBR^{utt} require that syllables are counted, either based on transcription, classification or in real time. CBR has become widespread, especially in babbling research on clinical risk groups (see study II for an

overview). CBR has been found to increase with infant age and does not seem to be affected by socioeconomic status (Lee et al., 2018; Oller et al., 1994).

Another continuous babbling measure is the Mean Babbling Level (Stoel-Gammon, 1989). In this analysis, utterances are given a complexity measure from 1 through 3 and a mean is calculated. For example, utterances at level 1 consists of single vowels, single consonants or consonant-vowel combinations in which the consonant is not a true consonant. In contrast, utterances at level 3 consist of two or more consonant-vowel combinations where the place and manner of the consonants are different.

In research on clinical risk groups, researchers are often interested in the timing of babbling onset. Different measures have been used to operationalize this onset. McGillion and colleagues (2017) identified the onset as the age at which an infant stably produces at least two different consonants. Schauwers and colleagues (2004) on the other hand used a multisyllabicity criterion; an infant was credited with babbling onset when they consistently and over sessions used multiple articulatory movements combined with phonation. A more common way of operationalizing the babbling onset is using the CBR together with a criterion for the canonical babbling onset, most often 0.15 (Bartl-Pokorny et al., 2022; Bass-Ringdahl, 2010; Belardi et al., 2017; Chapman et al., 2001; Iyer & Oller, 2008; Lieberman et al., 2019; Lohmander et al., 2017a; Lynch et al., 1995; Löfkvist et al., 2020; Nathani et al., 2007; Overby et al., 2020a; Overby et al., 2020b; Patten et al., 2014; Price et al., 2006; Willadsen & Albrechtsen, 2006; Willadsen et al., 2022). Thus, the onset of CB is credited when at least 15% of a child's production is canonical. Originally based on data from six children (Lynch et al., 1995), the 0.15 CBR criterion has, however, never been thoroughly evaluated.

2.3 SPEECH AND LANGUAGE DISORDER IN CHILDREN WITH NEUROLOGICAL DISABILITIES

2.3.1 On terminology

2.3.1.1 Speech disorder

Difficulties with speech production is common in children, both in children with disabilities and in children with otherwise typical development. While there has been quite a lot of debate regarding terminology, the umbrella term speech sound disorder (SSD) is now commonly used. In the early school age, SSD has a prevalence of 3–4 % (Eadie et al., 2015; Wren et al., 2016). Although SSD is common in neurological disabilities (ND), children with ND constitute a small minority of all children with SSD. Indeed, most research on speech sound disorder have been focused on so called idiopathic SSD, that is, SSD in children without other disabilities.

There are several different ways of classifying SSD subtypes (see Waring & Knight, 2013 for a discussion), but in children with ND, the distinction between motor speech disorders and non-motor speech disorders is the most relevant. Motor speech disorders in children include childhood dysarthria (defined by difficulties executing motor speech movements) and childhood apraxia of speech (CAS; defined by difficulties programming and planning speech

movements). A third category has also been suggested by Shriberg and colleagues: speech motor delay (Shriberg et al., 2019).

2.3.1.2 Language disorder

Many different terms have been used to refer to language difficulties in children, including, among many others, language impairment, language delay and language disorder. Efforts have often been made to distinguish between language difficulties with no known cause, and language difficulties co-ocurring with for example hearing loss, autism or genetic syndromes (Bishop, 2014). In a consensus project, the CATALISE consortium (Bishop et al., 2017) recommends the term "language disorder" for consistent language problems that significantly impact everyday life, regardless of whether other diagnoses are present. The term "language disorder associated with…" is further recommended for cases when the language disorder co-occurs with conditions such as hearing loss, genetic syndromes, autism or intellectual disability. When no such condition is present, the term "developmental language disorder" is recommended. The extension of the term language disorder to include children regardless of other diagnoses has not been entirely uncontroversial, especially when it comes to children with intellectual disability (ID) (Bishop et al., 2016).

In this thesis, "speech disorder" will be used for significant difficulties with the production of speech sounds. "Language disorder" will be used according to the CATALISE definition, unless specified otherwise. The term "speech/language disorder" will be used when it is unclear if a child has speech disorder, language disorder or a combination of the two, or when the distinction is irrelevant for the topic in question. Two related terms will also be used: "communication difficulties" and "communication problems" . These terms will be used interchangeably to refer to difficulties communicating in everyday life, regardless of the cause of these difficulties.

2.3.1.3 Neurological disabilities

One challenge with reading the research literature on speech and language disorder in neurological disabilities is that children with ND may be found under many different labels. One common way of labelling is using the medical diagnosis causing the child's disability, for example cerebral palsy (CP) or Down syndrome (DS). In addition to DS, there is an abundance of genetic syndromes that may cause developmental disability, many of which are rare or very rare. Some rare genetic syndromes are well researched when it comes to speech and language (see for example Williams syndrome), but many are less so. Thus, finding relevant and reliable information on the possible speech and language development for a young child with a rare genetic syndrome is not always easy.

Intellectual disability (ID), also referred to as intellectual development disorder, is defined as severe difficulties with intellectual abilities (corresponding to a result lower than two standard deviations below the mean on standardized tests) together with severe difficulties in adaptive behavior (*Diagnostic and statistical manual of mental disorders : DSM-5-TR*, 2022). The difficulties need to have been present since childhood in order to be classified as ID. The

severity of intellectual disability is often specified as mild, moderate, severe or profound (*Diagnostic and statistical manual of mental disorders : DSM-5-TR*, 2022). ID may be diagnosed in children with an underlying etiology (such as CP or a genetic syndrome) or in isolation (more common in mild ID). As ID is common in children with neurological disability, it constitutes another label under which children with ND may be found in research.

Nowdays, it is less common to conduct research on groups of children with ND with a variety of different medical diagnoses (as in this thesis). Earlier examples of this include research by Yoder, Warren, and colleagues and Brady and colleagues, examining children on the prelinguistic level of language development (see for example Brady et al., 2004; Yoder & Warren, 1998, 2001; Yoder et al., 1998). Included children had developmental delay and a variety of medical diagnoses and are referred to as "children with developmental disabilities" or "children with developmental delay" in different publications. To conduct studies on the speech and language characteristics on children with ID in general is rare (but see Murfett et al., 2008 and Loveall et al., 2016).

Two more labels that include children with ND that are of common use today will be mentioned. "Children with complex communication needs" is a term commonly used in research on augmentative and alternative communication (AAC) (see for example Light & Drager, 2007; Light & McNaughton, 2015). Children with complex communication needs are children who are in need of AAC, which include many children with ND. "Profound intellectual and multiple disabilities" (PIMD) is a term for severe disabilities characterized by co-occuring profound intellectual disability and profound neuromotor dysfunction, leading to functioning at a pre-symbolic level and at high risk for medical complications (Nakken & Vlaskamp, 2007). Thus, children with PIMD can be said to constitute a subgroup of children with ND.

In the following, previous research on speech and language in ND are summarized for twelve different medical diagnoses. This includes a review of previous research in DS and CP as well as short summaries of speech and language in ten rare genetic syndromes. Although not exhaustive, the presentation of the rare genetic symptoms is meant to give an overview of the variety of presentations in the category of children with ND. Descriptions of speech and language in children with PIMD or complex communication needs will not be explicitly covered, nor will the presentation include research on speech and language in children with autism.

2.3.2 Cerebral palsy

Cerebral palsy is a group of disorders of movement and posture, due to damage in the developing brain before 2 years of age. It is a heterogenous disorder, both in presentation, severity and in underlying etiologies (Rosenbaum et al., 2007). It has a prevalence around 2 in 1000 live births in western Europe (Himmelmann & Uvebrant, 2018; Surveillance of Cerebral Palsy in Europe (SCPE), 2002). CP is usually classified according to type of motor disorder (spastic, dyskinetic or ataxic, with spastic CP commonly divided into unilateral and bilateral

presentations) and according to functional gross motor ability (Cans, 2000). CP is non-progressive, although symptoms may change over the life course. Nowadays, it is emphasized that CP is not solely a motor disability, but is often accompanied by, for example, difficulties with communication (Rosenbaum et al., 2007).

Communication problems in CP may be due both to a speech disorder (caused by difficulties in motor speech control) and to cognitive difficulties (e.g., a language disorder), but speech disorders have been more thoroughly studied (Hustad et al., 2010). Speech disorder is common in CP, but the exact prevalence varies between studies; figures between 36% and 90% have been reported (Mei et al., 2014; Parkes et al., 2010). The variation is likely due to different age spans used in different studies, differences in participant sampling, and, more importantly, how speech disorder is defined and measured. The highest prevalence figures seem to come from studies where specialists assess presence of motor speech disorders in pre-school-aged children (Hustad et al., 2017; Mei et al., 2014), whereas the use of register data and/or data reported by physicians seems to render lower prevalence figures (Nordberg et al., 2013; Parkes et al., 2010). When it comes to language disorder, Mei and colleagues (2016) reported a prevalence of 62% in 5–6-year-olds. The prevalence of communication difficulties have also been examined in children with CP. Kristoffersson and colleagues (2020) examined the prevalence of communication disorder as measured with the Communication Function Classification Scale using Swedish registry data from children aged 0-18. Forty-five percent of participants was rated as effective communicators in all environments, and thus 55% had some type of communication difficulty. Pennington and colleagues (2020)used the same measure on data from five-year-olds with CP. Here, 82% were rated as having some kind of communication difficulty.

Symptoms of speech disorder identified in children with CP include reduced intelligibility (Chen et al., 2018; Hustad et al., 2019; Hustad et al., 2012), deviant consonant articulation (Nordberg et al., 2014; Workinger & Kent, 1991), reduced coordination between articulators (Nip, 2017), reduced vowel space (Chen et al., 2018), increased pause duration (Kuschmann & Lowit, 2020) and short phrases (Kuschmann & Neill, 2015). When it comes to types of speech disorder, dysarthria is the most common type, but childhood apraxia of speech and non-motor speech sound disorders have also been reported (Mei et al., 2020).

When it comes to symptoms of language disorder, Mei and colleagues (2016) reported that children with language disorder mostly had difficulties across language subdomains and with both receptive and expressive language. Other findings on language in CP include difficulties with sentence comprehension (Geytenbeek et al., 2015) and narrative ability (Nordberg et al., 2015).

Communication abilities in children with CP are strongly affected by motor and cognitive functioning. For example, degree of gross motor disability has been shown to be associated with concurrent expressive language skills (Choi et al., 2017) and to presence of speech motor impairment (Nordberg et al., 2013). Classification of manual ability has also been shown to be highly correlated to different classifications of communication (Choi et al., 2018). When it

comes to cognitive functioning, non-verbal cognitive ability is strongly correlated to receptive language (Soriano & Hustad, 2020). Presence of intellectual disability has been shown to significantly affect receptive language (Vos et al., 2014), as well as everyday communication (Choi et al., 2018). Presence of intellectual disability and poorer intellectual functioning are also associated with articulation difficulties in children with CP (Choi et al., 2017; Nordberg et al., 2014).

Communication in CP also seems to be affected by type of brain lesion, especially when it comes to expressive abilities. Expressive language ability has been shown to differ between types of brain lesion (Choi et al., 2017), and the distribution of lesion types has been shown to be different in groups with and without speech disorder (Nordberg et al., 2013). Peri-ventricular white matter lesions tend to be associated with more functional speech and communication, whereas basal gangliga lesions are associated with being non-verbal and having poorer communication (Himmelmann et al., 2013).

Recent years have seen quite a lot of longitudinal studies on speech and language development in CP. In general, speech and intelligibility improves with time, at least until the age of 10 (Hustad et al., 2019; Long et al., 2022). The development of intelligibility seems to vary between subgroups, with children with speech and language disorder being less intelligible than children with only speech disorder or children without speech and language disorder (Mahr et al., 2020).

There are indications that some children with CP may be initially delayed in their speech and language development and catch up on their peers during the pre-school years. In a longitudinal study by Hustad and colleagues, only 15% were established talkers at age 2, but 27% had typical speech at age 4 (Hustad et al., 2017). Thus, some children who have difficulties at age 2 can be expected to have outgrown their difficulties at age 4. On the other hand, children who were classified as "non-talking" at age 2 all had speech disorder at age 4, and 73% had anarthria (Hustad et al., 2017). In another study, Hustad and colleagues (2018) found two different patterns in a longitudinal study of 84 children. One group of participants, most of whom were non-speaking due to anarthria, had significantly lower language comprehension than age norms at both age 2 and age 4 years and in addition, a significantly slower rate of development. In the other group, language comprehension was significantly lower than age norms at 2 years, but not at 4 years. The rate of development between 2 and 4 years was significantly higher than age norms. Thus, it seems like children that do not have anarthria to some extent can "catch up" when it comes to development of language comprehension.

2.3.3 Down syndrome

Down syndrome is the most common chromosomal cause of intellectual disability and has a global incidence of 1/800 live births (Bull, 2020). The intellectual disability is often moderate, but ranging from mild to severe. Down syndrome is associated with increased risk for many medical conditions, including congenital heart disease, otitis media with effusion, thyroid abnormalities and dementia. Other characteristics of Down syndrome include general

hypotonicity and differences in oral anatomy including a small mid-face, a relatively large tongue and a high palate (Kent & Vorperian, 2013).

It is well known through both research and clinical experience that DS is associated with considerable challenges when it comes to speech and language abilities. People with DS usually present with a characteristic speech and language profile; reduced intelligibility and difficulties with expressive language and grammar are well documented (Abbeduto et al., 2007; Kent & Vorperian, 2013).

When it comes to language abilities, expressive language is generally found to be more impaired than receptive language, and language abilities in general more impaired than nonverbal cognitive abilities (see for example Cleland et al., 2010). The syntactic difficulties in DS has been labelled specific, as they are more prominent than in other NDs (Abbeduto et al., 2007). Relative strengths in the DS language profile include imitation, use of gestures and receptive vocabulary, particularly at older ages (Abbeduto et al., 2007; Næss et al., 2011).

Quite a lot of research effort has been put into whether language difficulties in DS constitute a delayed or disordered development. Using parent-reported data, Polisenka and Kapalkova (2014) conclude that children with DS present with the same gap between expressive and receptive vocabulary as seen in younger children with similar vocabulary size. They also used the same semantic categories and the same level of grammar, suggesting a delay rather than a disorder. Berglund and colleagues (2001) used a similar method and came to similar conclusions. In their study, children with DS were found to acquire grammatical markers in the same order as typically developing children and to combine words at the same level of lexical development. Using both parent-report and analysis of language use in parent-child interaction, Zampini & D'Odorico (2011) found support for a disorder hypothesis. Children with DS had a less complex vocabulary than typically developing children matched for vocabulary size and mental age, with fewer adverbs and fewer multi-word utterances. Children with DS also seemed to use established words more sparsely. In summary, children with DS seem to follow the same trajectory of language development as children with typical development but with a significant delay, while some also present with specific disorders. There are qualitative differences in the frequency of use of words as well as a specific difficulty with different types of grammatical markers. Speech and language ability in children with DS can therefore be considered both delayed and disordered (Abbeduto et al., 2007; Kent & Vorperian, 2013).

As mentioned above, speech production is a major challenge for many individuals with Down syndrome. As an example, Cleland and colleagues (2010) examined 15 children and adolescents (9–18 years of age) and found that a majority performed below the level of three-year-olds on a speech production test. It has been suggested that the speech disorder in DS is in fact due to dysarthria or CAS. Rupela and colleagues (2016) set out to investigate this in a study of six children aged 3–8 years. The participating children’s speech was analysed according to checklists of symptoms of CAS, symptoms of dysarthria and symptoms occurring in both CAS and dysarthria. The authors concluded that the participants with DS showed signs of both childhood apraxia of speech, dysarthria and unspecified motor-speech disorder. Thus,

children with DS often have a motor speech disorder in addition to a language disorder, but there is a huge variation in severity and speech characteristics between individuals.

Differences in orofacial anatomy, intellectual disability and hearing impairments due to otitis media with effusion are all common characteristics of DS. However, neither difficulties with syntax nor speech difficulties can be fully explained by these factors (Abbeduto et al., 2007; Kent & Vorperian, 2013). Verbal short-term memory (measured with word- or digit-span tasks) has been shown to be a specific difficulty in DS (Jarrold et al.; Næss et al., 2011), and might contribute to speech and language difficulties.

In general, studies of speech and language abilities in Down syndrome have been cross-sectional, and/or have examined children in a wide age span, but there are some important exceptions. Sokol and Fey (2013) compared the development of 24–33 month-old children, with DS or other NDs on different speech related measures. At study intake, children with DS had better results on all of these measures. At follow up 18 months later, the children with other types of ND had significantly better results on all measures. Thus, children with DS seem to have a slower speech development during the third and fourth year of life compared to children with other types of neurological disabilities. Naess and colleagues (Næss et al., 2021; Næss et al., 2015) examined the longitudinal language development of children with DS between 6 and 8 years of age compared to younger typically developing controls matched for non-verbal mental ability. Children with Down syndrome were outperformed by the matched typically developing children on all measures over all time points, except for receptive vocabulary at age 6. Both groups developed their language abilities over time, but changes were much greater in children with typical development. There was also a difference between groups when it comes to predictors of later language skills. Vocabulary was a predictor of grammar in typically developing children, but not in children with Down syndrome (Næss et al., 2015). Expressive vocabulary was predicted by home literacy, auditory memory and receptive vocabulary for both groups, but for the children with DS, oral motor skills and phonological memory were additional predictors (Næss et al., 2021).

2.3.4 Other genetic syndromes

The selection of rare genetic syndromes presented here aimed at including the most prevalent genetic syndromes, with the exception of sex chromosome disorders. All included syndromes, although occurring in less than 5/10 000 (Socialstyrelsen, 2022), are relatively often encountered in habilitation and early intervention services. Although each syndrome comes with its own unique phenotype, there are also many common features. The ten genetic syndromes often cause developmental delay or intellectual disability and affect organs of the body, often causing multiple health issues. Another common feature is the variability of presentation within the same genetic syndrome.

2.3.4.1 Genetic syndromes associated with no speech

Examples of genetic syndromes associated with no or very little speech are Rett syndrome, Trisomy 13 and 18, and Angelman syndrome. People with these syndromes often communicate

non-verbally, using eye gaze, vocalizations, facial expressions or body movements. Use of gestures are common in Trisomy 13/18 and Angelmann syndrome and AAC use is reported in Rett syndrome and Angelmann syndrome (Bartolotta et al., 2010; Braddock et al., 2012; Pearson et al., 2019; Wandin et al., 2015).

Mainly seen in women and girls, Rett syndrome is characterized by a regression in development after 6 months of age, especially when it comes to spoken language and manual ability. Partial or complete loss of speech is part of the diagnostic criteria (Neul et al., 2010). In addition, the syndrome leads to movement disorder and stereotypic hand movements. Approximately 77% of people with Rett syndrome are reported to have used words before the regression, but only 21% used words after the regression (Urbanowicz et al., 2015). A time delay when responding to stimuli is reported in many cases, as well as limb apraxia affecting communication (Bartolotta et al., 2010).

Trisomy 13 and Trisomy 18 are two syndromes associated with a very high fetal and infant mortality (Meyer et al., 2016). Individuals with Trisomy 13 and Trisomy 18 often have severe medical complications and severe disabilities, including severe communication difficulties. Most individuals use no or only a couple of spoken words. Vocabulary comprehension is a relative strength, although severe language comprehension difficulties are present (Braddock et al., 2012).

Angelman syndrome is characterized by intellectual disability that is often severe, movement and balance disorder and a distinct behavioral profile with frequent laughing/smiling and excitability. Speech/language disorder is a consistent feature in Angelman syndrome, with most individuals using no or very few spoken words (Williams et al., 2006). Although spoken language is rare, people with Angelman syndrome use a wide variety of communicative behaviors, especially non-symbolic communication (Pearson et al., 2019).

2.3.4.2 Genetic syndromes associated with no speech or speech/language disorder

There are also genetic syndromes which result in speech and language abilities that are highly variable between individuals, with some presenting with no speech, most with varying degrees of speech and language disorder and a few with typical presentations. Examples of these types of syndromes are Monosomy 1p36 deletion syndrome, Prader-Willi syndrome and Fragile X syndrome.

Monosomy 1p36 deletion syndrome is the most common terminal deletion in humans and comes with symptoms such as intellectual disability, hearing loss, seizures, growth impairment and distinct facial features. Speech delays are present in 98% of individuals with the syndrome and a mean onset of spoken language at 4–5 years of age has been reported (Brazil et al., 2014; Gajecka et al., 2007). In a survey of 40 adolescents and adults with 1p36 deletion, 44% were reported to use speech and 38% used speech in sentences. Use of manual sign and aided AAC was also common (Brazil et al., 2014).

Prader-Willi syndrome (PWS) is characterized by infant hypotonia, hypogonadism and short stature. People with PWS often present with intellectual disability (mostly in the mild range) or borderline intellectual functioning. Failure to thrive in infancy later develop into hyperphagia during childhood. PWS is associated with behavioral symptoms such as rigidity and compulsiveness, and autism and ADHD are common (Cassidy et al., 2012). When it comes to speech and language, presentations vary from non-verbal presentations to abilities in the normal range (Lewis et al., 2002), although mean results on language tests have been found to be in the very low range (Dimitropoulos et al., 2013). Examination of children and adults with PWS has shown a high occurrence of speech disorder, although variability was large (from mild speech disorder to severe) and results were higher in adulthood. Oral motor difficulties, hypernasality and atypical voice pitch were also reported (Lewis et al., 2002).

Resulting from a mutation on the X chromosome, Fragile X syndrome (FXS) is the most common heritable cause of intellectual disability (Mazzocco, 2000). Symptoms of FXS are variable, ranging from severe intellectual disability and autism to normal IQ (Garber et al., 2008). Girls and women, having two x-chromsomes, in general have less severe symptoms (Mazzocco, 2000). A proportion of individuals with FXS do not use speech to communicate, but there are different reports on how large this proportion is (see for example Abbeduto et al., 2016; Finestack et al., 2009; Levy et al., 2006). When it comes to language, people with FXS show impaired ability accross language domains. Language abilities are often are in line with those of typically developing children of the same mental age, but the gap compared to peers of the same chronological age become more prominent as children grow older (Finestack et al., 2009; Hoffmann et al., 2020). Difficulties larger than expected from mental age can be found in the pragmatic language domain, such as providing necessary information and the use of repetitive language. Speech intelligibility is often in line with the expected for mental age. Girls and women with FXS tend to have stronger language skills than boys and men, as do individuals without a co-occurring diagnosis of autism (Finestack et al., 2009).

2.3.4.3 Genetic syndromes associated with speech/language disorder

In a third group of rare genetic syndromes, non-verbal presentations are rare, but speech and language disorder are common. Examples in this group is 22q11 deletion syndrome, Sotos syndrome, Williams syndrome and Noonan syndrome.

The prevalence of speech/language disorder in 22q11 deletion syndrome is approximately 95% and the difficulties are complex in nature. Speech/language development is affected by co-existing conditions common in the syndrome, such as cleft palate, velopharyngeal dysfunction, otits media with effusion, developmental delay, hypotonicity and psychological and psychiatric disorders. Especially in early childhood, expressive language is more affected than receptive language. In school-age, children often have difficulties with grammar, vocabulary and pragmatics. Speech disorders are common and may be of different types; both motor-based and non-motor-based disorders occur, with or without concommittant velopharyngeal dysfunction (Solot et al., 2019).

Sotos syndrome has three main features: characteristic facial appearance, childhood overgrowth and intellectual disability (usually in the mild–moderate range) (Tatton-Brown & Rahman, 2007). Language abilities have been reported to be in line with intellectual level (Finegan et al., 1994), but stronger verbal than non-verbal cognitive functions (both measured by a cognitive test battery) have been reported (Lane et al., 2019a). Still, a majority of studied individuals with Sotos syndrome have reported communication difficulties (Lane et al., 2019b).

Characteristics of Noonan syndrome include, among others, distinct facial and musculoskeletal features, cardiac issues, short stature and feeding difficulties (Romano et al., 2010). Although the exact prevalence is not fully understood, speech, language and communication disorders seem to be more common in Noonan syndrome than typical development, although by no means universal. In a study by Pierpont and colleagues (2010) the prevalence of language disorder was approximately 30%, the prevalence of social-pragmatic problems approximately 40% and of speech disorder approximately 20%. Most individuals with Noonan syndrome show results on language tests that are slightly below the normative mean. Language abilities are highly correlated to non-verbal cognition, with no evidence of "specific" language disorder (Pierpont et al., 2010).

Williams syndrome is a rare syndrome which has attracted much research interest due to its unusual behavioral presentation, with language abilities considered to be normal despite significant intellectual disability. The somatic profile of the syndrome includes heart anomalies and distinctive facial features. Williams syndrome is often associated with mild intellectual disability, although there are large individual variations. Cognitively, concrete language tasks and verbal short-term memory are relative strengths, whereas visuospatial abilitiy is a weakness (Mervis & John, 2010). Despite the initial notion that language abilities were "spared" in Williams syndrome, researchers now agree that the syndrome is associated with language disorder, albeit with an uneven profile. Concrete vocabulary, both receptive and expressive, is a particular strength. Vocabulary for relational concepts, however, is a weakness (Mervis & John, 2008). Although people with Williams syndrome are often described as very socially interested, pragmatic difficulties are a part of the language profile (Mervis & John, 2010).

2.3.5 Co-existing conditions

Co-morbidity and co-existing conditions are very common in ND. Intellectual disability, has already been discussed as a prominent feature of Down syndrome as well as many other genetic syndromes. ID is also common in cerebral palsy, with a Swedish population-based study reporting a 51% prevalence (Påhlman et al., 2021). Other common co-existing conditions that affect the everyday functioning of children with ND include autism, ADHD, visual impairment and hearing loss. Here, examples of how children with DS and CP are affected by these co-existing conditions will be given.

Autism and ADHD are common in children with ND, but not always diagnosed. Swedish prevalence studies using team-based evaluations estimate the prevalence of autism to 30% in children with CP and 42% in children with DS. For ADHD, reported prevalence figures are 30% for CP and 34% for DS (Oxelgren et al., 2017; Påhlman et al., 2021).

Prevalence of visual impairment and hearing loss varies in the ND group based on the underlying etiology. In Down syndrome, hearing loss has been reported in up to 84% of children, with conductive hearing loss being the most common presentation (Bull, 2020; Kreicher et al., 2018). In children with CP on the other hand, prevalence of hearing loss is much lower, 4–13%, with sensorineural hearing loss being the most common (Reid et al., 2011). Visual impairments are common in children with CP, especially cerebral visual impairment and/or difficulties with visual perception (Guzzetta et al., 2001). In Down syndrome, severe refractive errors and cataract are reported to have a prevalence of 50% and 15%, respectively (Bull & the Committee on Genetics, 2011).

2.4 BABBLING IN NEUROLOGICAL DISABILITIES

From the above, it is clear that speech and language are often affected in children with neurological disabilities. Despite this, relatively few studies have systematically examined babbling in children with ND. As in babbling research in general, research in infant vocalizations in ND has used varied methodology and targeted different ages, rendering results challenging to synthesize (Lang et al., 2019; Roche et al., 2018). There are, however, indications that babbling is affected in ND. In Oller and collagues' study on telephone screening of canonical babbling (Oller et al., 1998), confirmed or suspected neurological disability was over-represented among the children who had not reached the canonical babbling stage. Suspected or confirmed diagnoses among the group of children not in the canonical babbling stage included CP and different genetic syndromes. Some of the disabilities among the non-canonical group had not been diagnosed prior to the telephone screening. Another early study by Oller ang colleagues (Oller & Seibert, 1988) examined babbling in children with what today would be called intellectual disability. In addition, the participants had a wide variety of medical conditions, including for example Down syndrome and microcephalia. The participants were 17 to 62 months old, had a mean mental age of 17 months and where all in the pre-linguistic stage of development. Out of the 36 participating children, 29 had entered the CB stage. The cross-sectional methodology of the study did not permit any conclusions as to whether the 29 canonical participants had entered the CB stage in a timely manner. The study is, however, one of the first examples of CB being described as necessary, but not sufficient, to the development of speech in children with ND.

Two studies have examined babbling in children with CP or suspected CP, both indicating delays compared to typically developing children. Levin (1999), studying eight infants, reported a delayed CB onset and small phonetic repertoires with only monosyllables. Ward and colleagues (2022), examining 18 children longitudinally using parent interviews, reported no difference compared to typically developing infants at 6 months, but at 9 and 12 months, indicating an increased babbling delay with time. Both studies included infants with more severe gross motor symptoms, compared to the whole CP continuum.

Another diagnosis where deviant pre-linguistic vocalizations have been reported is Rett syndrome. Although some children with Rett syndrome do reach the CB stage in a timely manner (Bartl-Pokorny et al., 2022), and use of CB can be seen in some children pre-regression

(Lang et al., 2019), many atypicalities have been reported. This includes lower CBR and less complex syllable shapes but also frequent ingressive vocalizations and high-pitched cries (Bartl-Pokorny et al., 2022; Marschik et al., 2012).

Delayed CB onset has also been reported in William's syndrome (Masataka, 2001) and Fragile X syndrome (Belardi et al., 2017). In the latter case, however, contradictory reports finding no differences compared to babbling in children with typical development also exist (Hamrick et al., 2019).

Down syndrome constitutes another example of small or no differences in babbling compared to typically developing children. As Kent and Vorperian state in their systematic review (Kent & Vorperian, 2013), any difference between babbling in children with DS and children with typical development is smaller than would be expected, given the severe speech difficulties that are associated with DS. Reported differences include a delayed (by approximately 2 months) and less stable CB onset in DS (Lynch et al., 1995). Babbling in children with Down syndrome has however been reported to be qualitatively similar to babbling in children with typical development when it comes to the proportion of reduplicated and variegated babbling (Smith & Stoel-Gammon, 1996). Steffens and colleagues (1992) found significant differences in CBR at 16 months of age, but at the other ages examined no difference was seen compared to children without DS. Indeed, the CB status of children with Down syndrome has been taken as an argument for the universality of canonical babbling as a milestone.

Even more sparse than the research on babbling in ND, is the research on babbling as a predictor for speech and language in ND. Previous studies have exhibited large variation when it comes to participant diagnoses, babbling measures and predicted variables, but some associations between canonical babbling and later language and communication measures have been reported (see table 1).

Table 1. Summary of previous studies on babbling in children with neurological disabilities.

Reference	Diagnosis	Babbling variable	Predicted variable
Lynch et al., 1995	Down syndrome	Age at parent reported CB onset	Social communicative functioning at 27 months
Masataka, 2001	William's syndrome	Age at CB onset	Age at first word
Hamrick et al., 2019	Fragile X syndrome	Use of canonical syllables at 9 months	Results on receptive and expressive language tests at 24 months.

2.5 HABILITATION SERVICES IN SWEDEN

The habilitation services in Sweden is a multi-professional, team-based type of service for children, youth and adults with disabilities, provided by the local regions (county councils). Here, a short description of habilitation services for children will be provided. Aimed at children with physical or cognitive disabilities, habilitation services are complementary to other health care services (Ylvén, 2013). Although there is regional variation, the services are commonly targeted towards children with movement disorder (e.g., CP), intellectual disability and autism. Thus, children with ND (according to the definition used in this thesis) are among the groups entitled to support from the habilitation services. Regions also often provide specific habilitation services for children with hearing loss or visual impairment. Professions employed in the habilitation services are subject to regional variation, but often include physiotherapists, occupational therapists, psychologists, social workers, special educators and speech-language pathologists. Physicians, nurses and dieticians may also be part of the team. Interventions are aimed at enhancing participation in daily activities, develop the child's abilities, improve child well-being and provide support for parents (Wettergren et al., 2016). Collaboration with for example pre-schools, schools, social welfare services and psychiatry is common. Habilitation services are regulated under the Swedish Health and Medical Service Act (*Hälso- och sjukvårdslag* SFS 2017:30). Regions are obliged to provide habilitation services, and the patients are entitled to an individual plan for their services. As with all publicly funded health care in Sweden, the local regions have considerable freedom to decide how to organize the habilitation services (Wettergren et al., 2016). Thus, there are no national regulations regarding the content or frequency of services.

2.6 TREATMENT FOR SPEECH AND LANGUAGE DISORDER IN NEUROLOGICAL DISABILITIES

A variety of different interventions for speech, language and communication have been evaluated in neurological disability, including different versions of direct speech and language therapy, parent-implemented therapy and AAC interventions (see for example Akamoglu & Meadan, 2018; Gevarter & Zamora, 2018; Nordahl-Hansen et al., 2019; Simacek et al., 2018). In Sweden, there are national recommendations for evidence-based practice within the habilitation services (Backman et al., 2015; Eberhart et al., 2011)[2]. For preschoolers on a prelinguistic or early linguistic level of development a combination of direct and indirect intervention is recommended, focusing on responsive interaction, joint attention, imitation and use of symbols to communicate. Intervention shall be delivered in naturalistic contexts and multi-modal AAC shall be introduced early (Eberhart et al., 2011). For school-age children, adolescents and adults recommendations include AAC interventions (including picture exchange communication system, PECS) and direct language training (Backman et al., 2015).

[2] The recommendations include all diagnoses qualifying for habilitation services. Recommendations for people with autism are however not included in this summary.

In this thesis, focus will be on intervention for speech/language disorder in CP and in the following, previous research on this topic will be presented.

2.6.1 Treatment for speech and language disorder in CP

As in many ND diagnoses, children with CP and speech and language disorder can benefit from different types of intervention, including AAC, direct training by an SLP or indirect training delivered for example by a parent under the supervision of an SLP. AAC can be beneficial for many children with CP and is crucial for individuals who cannot meet their communicative needs via speech (Hustad et al., 2012). However, this section will focus on research regarding non-AAC interventions for children with CP. Several systematic reviews regarding intervention for speech and language disorder in CP (and related patient groups) have been published (see below). Generally, results have revealed a lack of high quality studies in the field.

2.6.1.1 Direct intervention

A Cochrane review found no randomized controlled trials examining the effect of intervention for dysarthria acquired before the age of 3 years (Pennington et al., 2018). Published studies on direct intervention typically include few participants. Methods that have shown promise include Systems approach (Pennington et al., 2010) and Lee Silverman Voice Treatment (Fox & Boliek, 2012). These methods focus on respiratory control and phonation. Small studies have shown effects in the form of increased intelligibility (Boliek & Fox, 2017; Pennington et al., 2010; Pennington et al., 2013), changes in acoustic measures of speech (Boliek & Fox, 2017), increased communicative participation (Pennington et al., 2013) and speech judged as better by listeners (Boliek & Fox, 2017; Fox & Boliek, 2012). Maintenance effects are, however, still unclear for both methods. Other direct interventions that have shown promise in small studies on children with CP include two methods focusing on articulation: Prompts for restructuring oral muscular phonetic targets (Ward et al., 2014) and visual biofeedback from electropalatography (Nordberg et al., 2011).

2.6.1.2 Indirect intervention

One disadvantage of the direct intervention methods described above, is that they are difficult to implement in young children, as the demands are high when it comes to child cooperation and motivation. For young children (or participants on the developmental level of young children), indirect approaches are therefore often preferred. The goal is usually to promote better interaction between the child and a caretaker. As with direct therapy, studies are often small and lacking in quality (Chorna et al., 2017; Pennington et al., 2018; Pennington et al., 2004). In a Cochrane review (Pennington et al., 2018), including children with CP and other motor disabilities, two randomized controlled trials were found. These two studies showed that parents became more responsive to their children, but child communication did not improve. In a systematic review on training for communication partners of children with CP, (Pennington et al., 2004) all included studies reported effect on the communication partners, but changes in child communication were not always reported

Thus, even though parent training can change the communication of the parent, it is not clear whether this also leads to improved communication ability for the child with CP. Furthermore, even when the child's communication improves, it does not automatically lead to improvements in the child's speech or language abilities. Although a change in communication might be an appropriate goal for a specific child, meeting goals at the level of speech/language function seems to require other, more specific approaches.

3 RATIONALE FOR THE THESIS

As we have seen, babbling is a precursor to speech that has rendered a lot of research interest, but the fact that methods and measures have not been thoroughly validated means that this research may be hard to interpret. CBR is a common babbling measure in clinical babbling research, but it is time-consuming to use and the criterion for when a child is considered to be in the CB stage is based on six children and has not been validated after its original introduction. In this thesis, a simplified version of the CBR measure (CBR^{UTTER}) is used, which, if proven valid, may simplify the processes in clinical babbling research.

The Swedish habilitation services aim to modify the consequences of the disabilities for children with ND. Children with ND often have speech and language disorder, but much is still unknown when it comes to the longitudinal development of babbling, speech and language. Better understanding of this is needed in order to advice families, discover children in need of intervention and to design intervention programs.

Intervention for speech and language disorder in ND have generally not been sufficiently evaluated and SLP service delivery have not been examined within the context of the Swedish habilitation services. For children with CP in particular there is especially a lack of evaluation of interventions aimed at young children, focusing on child language goals.

4 RESEARCH AIMS

The general aims of the project were to

1. Validate different aspects of the canonical babbling ratio – including the 0.15 criterion for determining wheter a child is in the canonical babbling stage (study I and II),
2. Describe babbling and its association to later speech production in children with neurological disabilities (study I and III),
3. Describe the speech and language development in a group of children with neurological disabilities and explore its relation to SLP intervention (study III and IV), and
4. Evaluate a parent-implemented intervention for children with cerebral palsy and speech/language disorder (study V).

5 MATERIALS AND METHODS

This thesis consists of three different types of studies, using different data sets:

- Two methodological studies, using previously collected data from children with other diagnoses than ND (part of study I and study II)
- A three-part longitudinal study including children with different NDs (part of study I, study III and study IV)
- One intervention study including children with ND, specifically CP (Study V)

In this section, materials, procedures and statistic analysis will be described in short for the three types of studies. Thereafter, ethical considerations will be described for the project as a whole. An overview of the study designs can be found in table 2.

Table 2. Study designs.

Study	Design
I – part 1	Methodological validation study
I – part 2	Cross-sectional observation study with comparison group
II	Methodological validation study
III	Prospective, longitudinal observation study
IV	Prospective, longitudinal observation study
V	Single case A-B study with control behavior, repeated across four participants

5.1 METHODOLOGICAL STUDIES

In the methodological studies, a novel version of the CBR measure was examined – CBR^{UTTER}. Developed in the research group, CBR^{UTTER} is a simplified version of the CBR where the number of utterances containing canonical syllables is divided by the total number of utterances (see table 3 for a comparison of the formulae used for CBR^{UTTER} and for other versions of the CBR.) CBR^{UTTER} was validated in two different ways: using other versions of the CBR (study I) and using real-time babbling observation (study II). Furthermore, the 0.15 criterion for classifying children as being in the canonical babbling stage or not was examined, both concurrently and predictively (study II). The methodological studies used data from children with other diagnoses than ND, previously collected in our research group.

5.1.1 Materials

For the methodological studies, previously collected data were used (table 4). In study I, phonetic transcriptions of utterances from 12- and 18-month-old children with and without cleft palate were used. In study II, babbling, speech and language data had been collected within three different longitudinal studies focusing on babbling and early speech and language

development in typically developing children and children with different risk factors (otitis media with effusion, with or without cleft palate, sensorineural hearing loss treated with hearing aids and delayed babbling).

Table 3. Formulae for different versions of the CBR measure.

	Formula
CBR^{utt}	(Number of canonical syllables) / (Total number of utterances)
CBR^{syl}	(Number of canonical syllables) / (Total number of syllables)
CBR^{UTTER}	(Number of utterances containing canonical syllables) / (Total number of utterances)

Table 4. Data used in the methodological studies. Description of the participant groups, the number of participants providing data to each study, and the type of data used.

Group	Study I (part 1)	Study II	Data
Typically developing children (Lohmander et al., 2011)	11		Phonetically transcribed utterances at 12 months
	11		Phonetically transcribed utterances at 18 months
Children with cleft palate (Lohmander et al., 2011)	6		Phonetically transcribed utterances at 12 months
	10		Phonetically transcribed utterances at 18 months
Typically developing children (Persson et al., 2019)		21	CBR^{UTTER} based on counting of utterances at 10 months
			CB observation at 10 months
			Consonant production at 36 months[a]
			Parent-reported vocabulary at 30 months[b]
			Parent-reported vocabulary at 36 months[c]
Children with otitis media with effusion, with or without cleft palate (Lohmander et al., 2021)		9	CBR^{UTTER} based on counting of utterances at 10 months
			CB observation at 10 months
			Consonant production at 36 months[a]
			Parent-reported vocabulary at 30 months[b]
Children with hearing aid-treated sensorineural hearing loss (Persson et al., 2021a)		9	CBR^{UTTER} based on counting of utterances at 10 months
			CB observation at 10 months
			Consonant production at 36 months[a]
			Parent-reported vocabulary at 30 months[b]
			Parent-reported vocabulary at 36 months[c]
Children with babbling delay (Lieberman et al., 2022)		11	CBR^{UTTER} based on counting of utterances at 10 months
			CB observation at 10 months
			Consonant production at 36 months[a]
			Parent-reported vocabulary at 36 months[c]

*Note: **a** Swedish Articulation and Nasality Test, short version (Lohmander et al., 2015; Lohmander et al., 2017b). **b** Swedish Early Communicative Development Inventory – words & sentences (Berglund & Eriksson, 2000). **c** Swedish Communicative Development Inventory III (Eriksson, 2017)*

5.1.2 Procedures

In study I, three different versions of the CBR measure were calculated based on the phonetically transcribed utterances: the novel measure CBR^{UTTER}, together with the CBR^{syl} and CBR^{utt} (see table 3).

In study II, CBR^{UTTER} (based on the counting of utterances in audio-video recorded parent-child standardized interaction sessions) was compared to a babbling observation, where an experienced observer watched the same recordings and made a decision as to whether the child was in the canonical babbling stage or not (Lohmander et al., 2017a). In addition, CBR^{UTTER} at 10 months was related to the presence of speech/language difficulties at 30–36 months. Speech/language difficulties was defined as a result falling below age-norm levels at either the Swedish Articulation and Nasality Test (SVANTE; short version) (Lohmander et al., 2015; Lohmander et al., 2017b) or any of two parent-reported vocabulary measures (the Swedish Early Communicative Development Inventory: words and sentences – SECDI-w&s – at 30 months (Berglund & Eriksson, 2000) or the Swedish Communicative Development Inventory-III at 36 months (Eriksson, 2017)). SVANTE assessments had been phonetically transcribed and the percentage consonants correct (PCC) for the target sounds had been calculated. The PCC measure was originally developed for connected speech (Shriberg & Kwiatkowski, 1982), but in this case it was used on single words from the SVANTE, as has been done previously in research on children with cleft palate (Lohmander & Persson, 2008; Scherer et al., 2008). PCC values were then compared to SVANTE age norms for Swedish 3-year-olds (Lohmander et al., 2017b). A result at or below -1.5 standard deviations (SD) was categorized as indicative of speech difficulties. Difficulties with parent-reported vocabulary was defined as a result at or below the 10^{th} percentile, compared to Swedish age-norms. For SECDI-w&s, age-norms for 28 month old children were used, as norms for 30-month-olds are not available.

5.1.3 Reliability

For study I, reliability of transcriptions had been ensured in the original study (Lohmander et al., 2011). For study II, reliability assessments previously done in the context of the original studies were compiled and inter- and intrarater reliability were calculated for the 50 participants. Inter- and intra-rater reliability for the CBR^{UTTER} calculations were estimated using the Intra-class correlation coefficient. Inter-rater reliability was 0.87 (95% CI 0.72–0.99; two-way, random effects model, absolute agreement) and intra-rater reliability was 0.97 (95% CI 0.87–0.99; two-way, mixed effects model, absolute agreement). For the CB observation, intra-rater reliability was estimated using Cohen's kappa, $\kappa = 0.81$ (95% CI 0.46–1) and for the phonetic transcriptions of the SVANTE test, inter-transcriber reliability was estimated using percent agreement, point by point, 88%.

5.2 LONGITUDINAL STUDIES

5.2.1 Participants

For the longitudinal studies, data were collected from a total of 18 children with ND, recruited from habilitation centers in the Stockholm Region for this thesis project. In addition, data from typically developing controls were used for comparison in study I. Table 5 presents an overview of the participants in the longitudinal studies and figure 1 gives an overview of the recruitment and the attrition during the data collection process. Participants were originally recruited from seven different habilitation centers in the Stockholm Region. All children between 10 and 24 months receiving habilitations services, who had at least one parent with Swedish as their first language, and who used some sort of vocalizations were eligible to participate. Twenty-four families were asked to participate, six families declined.

Data were collected at three time points: at 1 year of age (T1; mean age 16 months, range 12–22 months), at 5 years of age (T2; mean age 5 years 1 month, range 4:11–5:4) and at 7 years of age (T3; mean age 7 years 7 months, range 7:4–7:10). Table 6 summarizes background information for the participants at this three time points. This background information is based on medical records (diagnosis, hearing at T2 and T3, intellectual disability), parent report (hearing at T1, visual impairment) and ratings by the author (motor ability). Due to attrition, information for some participants is only available for the first time point(s).

5.2.2 Procedures

A summary of the most important measures at each time point can be found in table 7. Because of the complex and sometimes severe disabilities the children had, the data collection was individualized to suit each child's abilities. Although many tests were included in the test battery, children only took tests that were deemed appropriate based on their developmental level and disabilities. The selection of tests was discussed with parents as needed. The participants were seen at a habilitation center or in their homes. The children's complex cognitive and motor disabilities required the testing procedure to be adapted to suit their needs. Adaptions included frequent pausing, visual support, adapted seating and allowing for other responses than finger pointing.

At T1, babbling was assessed based on audio-video recordings of parent-child interactions, using a standardized selection of toys. The recordings were 35–45 minutes long and parents were asked to play with their child as they normally would. Two types of assessments were done based on the parent-child recordings. First, a babbling observation was performed using a standardized observation form (Lohmander et al., 2017a). Among other things, the observer rated plosives and dental plosives as present or absent and marked all consonant sounds heard on a list of all Swedish consonant phonemes. Then, all utterances and all utterances containing canonical babbling were counted, and CBR^{UTTER} was calculated. Two different measures of babbled consonant repertoire were used in this thesis. In study I, the number of different consonants was used, whereas in study III the number of different *true* consonants was used, following McCune and Vihman (2001).

At T2 and T3, a test battery of different speech and language tests were used. Language tests were all scored according to the manual. Audio-video recordings from the test of consonant production were phonetically transcribed using semi-narrow transcription. Based on the transcriptions, PCC was calculated for the 30 target sounds in the single words part of the SVANTE test (Lohmander et al., 2017b). Results on speech and language tests were compared to Swedish age norms. Presence of speech and language disorder was defined using z-score criteria related to the prevalence of speech and language disorder. Based on a 4% prevalence of speech sound disorder (Wren et al., 2016), speech disorder was defined as a SVANTE PCC z score lower than -1.751. Based on a 7% prevalence of developmental language disorder (Norbury et al., 2016), language disorder was defined as a z score lower than -1.4 on at least one language test.

At T3, a rating of the participants communicative ability was done by the author using the communication activity scale from the Therapy Outcome Measures – AAC (TOM) (Enderby, 2014). In TOM, ratings are performed on a scale from 0 to 5, with 0 representing the worst possible presentation and 5 a normal presentation considering the person's age and cultural context. Each whole scale step is clearly defined and in addition, half points can be used to further distinguish between presentations. Data on SLP service delivery were collected through a parental questionnaire and through extraction of data from medical records.

Table 5. Participants in the longitudinal studies

	Study I (T1)	Study III (T2)	Study IV (T3)
Children with early diagnosed ND, recruited from habilitation centers	18	11	14
Typically developing children (Lohmander et al., 2011)	18	-	-

Figure 1. The data collection process in the longitudinal studies.

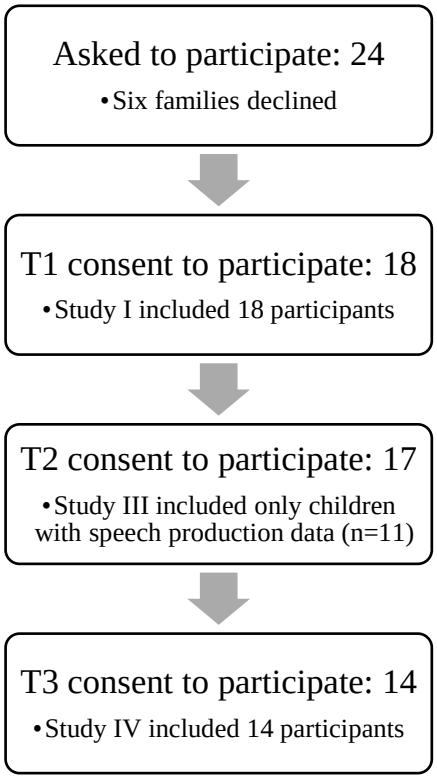


Table 6. Background characteristics of the participants in the longitudinal studies.

Participant	Sex	Diagnosis	Hearing T1	Hearing loss T2	Hearing loss T3	Visual impairment (T3)	ID (T3)	Motor ability (T3)
DS1	Boy	Down syndrome	Did not pass	Normal	Mild	Yes	Moderate	Walks with limitations
DS2	Girl	Down syndrome	Passed	Normal	Normal	Unknown	Mild	Walks without limitations
DS3	Boy	Down syndrome	Passed	Mild	Mild	No	Mild	Walks without limitations
DS4	Girl	Down syndrome	Did not pass	Mild				
DS5	Boy	Down syndrome	Passed	Mild	Mild	No	Mild	Walks with limitations
DS6	Boy	Down syndrome	Passed	Moderate	Moderate	No	Moderate	Walks with limitations
CP1	Boy	Cerebral palsy	Passed	Moderate	Mild	Yes	No ID	Transported in manual wheelchair
CP2	Girl	Cerebral palsy	Passed	Unknown	Normal	Yes	Mild	Walks with limitations
CP3	Girl	Cerebral palsy	Passed	Normal	Normal	No	No ID	Walks with limitations
CP4	Girl	Cerebral palsy	Passed	Normal	Normal	Yes	No ID	Walks with limitations

Participant	Sex	Diagnosis	Hearing T1	Hearing loss T2	Hearing loss T3	Visual impairment (T3)	ID (T3)	Motor ability (T3)
CP5	Boy	Cerebral palsy	Passed	Mild	Mild	Yes	Severe	Self-mobility with limitations
CP6	Girl	Cerebral palsy	Passed	Normal				
CD1	Boy	Chromosomal deletion	Passed	Normal	Normal	No	Moderate	Walks with limitations
CD2	Girl	Chromosomal deletion	Passed	Mild	Normal	Yes	No ID	Walks without limitations
CD3	Girl	Chromosomal deletion	Did not pass	Moderate	Moderate	Unknown	Severe	Walks with limitations
O1	Girl	No ethiology defined	Did not pass	Mild	Mild	Yes	Moderate	Walks with limitations
O2	Boy	Brain malformation	Did not pass	Moderately severe*				
O3	Girl	No ethiology defined	Passed					

***Note**: T1 = time point 1, T2 = time point 2, T3 = time point 3, ID = intellectual disability, CMV = cytomegalovirus infection, * = participant had bilateral cochlear implants at T2, results from an aided audiogram. Empty fields indicate that the child did not participate at that particular time point.*

***Hearing T1:** Results on the Ling Six Sounds Test (Smiley et al., 2004). "Passed" indicates a response to all six sounds, at any side and any distanc; **Hearing loss T2** and **Hearing loss T3**: Hearing level on the best ear classified according to World Health Organization (World Health Organization, 2021): Normal : <20 dB HL, Mild: 20 to <35 dB HL, Moderate: 35 to <50 dB HL, Moderately severe: 50 to <65 dB HL, Severe: 65 to <80 dB HL, Profound: 80 to <95 dB HL; **Visual impairment:** The presence of any parent-reported visual impairment regardless of type and severity.**ID:** Diagnosis according to ICD-10; **Motor ability:** Categories are based on the GMFCS (Palisano et al., 1997).*

Table 7 . Important measures used in the longitudinal studies.

Domain	Instrument / method	Measure	Study I (age 1)	Study II (age 5)	Study III (age 7)
Infant vocalizations	Babbling observation[a]	Use of plosives	X		
		Use of dental plosives[b]	X		
		N of different consonants	X		
		N of different true consonants		X	
	Counting of utterances containing CB	CBR^{UTTER}	X		
Consonant production	SVANTE, short version, phonetically transcribed[c]	PCC		X	X
Language comprehension	TROG-2[d]	Raw scores		X	X
Sentence recall	Subtest of the CELF-4[e]	Standard score		X	X
Communication ability	Ratings by SLP	TOM[f] level			X
Received SLP services	Parental questionnaire	Type and frequency of SLP services			X
	Data from medical records	Number of SLP sessions during the last year.			X

*Note: **a** (Lohmander et al., 2017a). **b** /t/ or /d/. **c**. Swedish Articulation and Nasality Test (Lohmander et al., 2015; Lohmander et al., 2017b). **d** Test for Reception of Grammar – 2 (Bishop & Garsell, 2009). **e** Clinical Evaluation of Language Fundamentals-4 (Semel et al., 2003). **f** Therapy Outcome Measures (Enderby, 2014).*

5.2.3 Reliability

Inter-rater reliability was assessed using percent agreement and intraclass correlation coefficients (single measures, absolute agreement).

5.2.3.1 Babbling observation

For study I, the babbling observation was re-assessed by a second observer. In addition, the first observer re-assessed five participants (28%). The inter-observer agreement was 89% for occurrence of plosives and 83% for occurrence of dental plosives. Intra-observer agreement was 100% for occurrence of plosives and 80% for occurrence of dental plosives. The intra-observer agreement was high for number of different consonants (ICC (two-way, mixed effects model) = 0.789, $p = 0.035$), but the inter-observer agreement was only fair (ICC (two-way, mixed effects model) = 0.401, $p < 0.0001$). Based on this lower reliability, it was decided to perform further reliability analyses for the measure number of different true consonants, used in study III. Five randomly selected participants were re-assessed and the agreement compared to the first observer was 76%.

5.2.3.2 Phonetic transcriptions

At both 5 and 7 years of age, phonetic transcriptions of the SVANTE target words were performed by two transcribers. Reliability was calculated based on the PCC values obtained from each transcriber. Inter-transcriber reliability was excellent at both ages (Age 5 ICC (two-way, random effects model) = 0.904, $p < 0.001$; age 7 ICC (two-way, random effects model) = 0.986, 95% CI 0.950–0.996).

5.3 INTERVENTION STUDY

The intervention used a single case A-B design, replicated across four participants. Unlike a case report, a single case A-B study is prospective, with a controlled introduction of the intervention and with repeated and pre-planned measurements of intervention effect. The use of single case design has been recommended in CP intervention research (Beckers et al., 2020; Pennington et al., 2004).

5.3.1 Participants

Four boys with CP (“Adam”, “Benjamin”, “Charlie” and “David”) participated in the intervention study. They were recruited from habilitation centres specifically for this study, and thus did not participate in the longitudinal study. They were 26–38 months of age at study intake and had spastic bilateral or spastic unilateral CP. Gross and fine motor disabilities varied from mild to moderate. Two participants were prematurely born (at 26 and 29 weeks of gestation). At study intake, participants were tested with the Bayley scales of infant-toddler development (Bayley et al., 2009). Index scores on the Bayley scales were below the means

for typically developing children on all scales (Cognition range 75–90, Language range 68–86, Motor range 55–85)[3].

5.3.2 Procedures

The study consisted of a baseline phase (A) and an intervention phase (B). During the baseline phase, parents received training in the intervention techniques and during the intervention phase they delivered the intervention at home for a minimum of 10-15 minutes daily. The intervention used a focused stimulation technique (Cleave & Fey, 1997); parents learned to repeat target words up to three times while maintaining a responsive interaction style and not placing any demands on the child to repeat the words. Parent coaching was provided by an SLP weekly during the intervention phase. Target and control words were probed throughout the baseline and intervention phases for a total of seven probes. Probes were audio-video recorded and assessed by the author and examiners blinded to the order of recordings as well as the target words for each child. All target and control words were understood but not produced by the children at study intake as per parent report, using the Swedish Early Communicative Development Inventory – words & gestures (SECDI-w&g) (Eriksson & Berglund, 1999). At the end of the baseline phase, the selected words were randomized to either control or target condition. The number of target and control words both varied between eight and ten over the four participants.

The primary outcome measure was the number of target words produced during probes. The number of produced control words during probes was used for comparison, intended to indicate whether any gains in produced words were restricted to the words included in the intervention or not. Parent-reported vocabulary (as measured by SECDI-w&g) at the end of intervention and at follow-up seven weeks later was used as a secondary outcome measure, intended to indicate whether generalization of vocabulary development had occurred.

5.3.3 Reliability

To ensure reliability of the assessments of the word probes, these were done by two different observers based on the recordings. Both observers watched the recordings of all word probes sessions and decided whether the words were produced or not. One observer was blinded to the allocation of target and control words, as well as to the chronological order of the recordings. The second observer (the author) had collected the data and was thus not blinded, but word probes were assessed at least six months after they were collected, to reduce recall bias. If the two observers disagreed on more than 10% of the decisions within one session, this session was re-assessed by a third blinded observer, and a majority decision was made on the words which the first two observers disagreed upon.

[3] The version of the Bailey scales used are not adapted for children with motor disabiltities and the results should therefore be interpreted with caution.

5.4 STATISTICAL ANALYSIS

An overview of the statistics used in the thesis can be found in table 8. Statistical analysis was perfomed using SPSS or R (R Core Team, 2019).

5.4.1 Methodological studies

In the methodological part of study I, the measure CBR^{UTTER} was compared to the traditionally used CBR^{syl} and CBR^{utt}. Pearson's correlation coefficient was used to examine the correlation between the measures and the intra-class correlation coefficient was used to check the agreement between the measures.

In the second methodological study (study II), a receiver-operating characteristics (ROC) curve was used together with area under the curve (AUC) statistics to examine the validity of the CBR measure compared to canonical babbling observation. An ROC curve is a graph of the sensitivity and specificity for a test across different criterion levels. Sensitivity and specificity is calculated in relation to a reference test, in this case canonical babbling observation. The AUC can thus be interpreted as the probability that a randomly chosen participant not in the CB stage according to observation has a lower CBR^{UTTER} than a randomly chosen participant in the CB stage according to observation. To identify the optimal CBR^{UTTER} criterion, positive likelihood ratios (LR+) for different possible criterion levels were examined. The LR+ indicates how much more likely a positive test is in a "sick" individual (an child not in the CB stage) , compared to a "healthy" individual (a child in the CB stage). Positive likelihood ratios were chosen for comparison instead of negative likelihood ratios, as the identification of children not in the CB stage often is of interest in clinical babbling studies, as opposed to the identification of children in the CB stage.

5.4.2 Longitudinal studies

In the longitudinal studies non-parametric tests were used, as the *n* was small and data were not normally distributed. Groups were compared using the Mann-Whitney U-test for continuous data and the Fisher's exact test for dichotomous data. In study IV, which was more exploratory, no inferential statistics were used. Instead, descriptive statistics was used to describe the development of speech and language and the SLP service delivery.

5.4.3 Intervention study

In the intervention study, the effect of the parent-implemented intervention was evaluated using visual analysis. Word probes for target and control words during the A and B phases were visualized in line graphs and results for target and control words were compared. Visual analysis is commonly used in the single case research, although there are also studies which use statistical analysis, for example overlap measures. These were considered for the intervention study, but not deemed appropriate as a rapid change between phases was not expected.

Table 8. Overview of statistics used in the different studies.

Study (type)	Aim for analysis	Statistics
I (M)	Comparison of different CBR measures	Pearson's correlation coefficient Intra-class correlation coefficient (two-way, mixed effects model, single measures, absolute agreement).
I (L)	Differences between children with ND and control data	Mann-Whitney U-test Fisher's exact test
II (M)	Validity of the CBR compared to CB observation	Receiver-operating characteristics curve with Area under the curve statistics
II (M)	Definition of CBR criterion for being in the CB stage	Comparison of positive likelihood ratios and sensitivity
II (M)	Comparison of different CBR criterion levels	Sensitivity and specificity
III (L)	Correlations between T1 and T2 measures	Spearman rank correlation
IV (L)	Development between 5 and 7 years of age	Descriptive statistics
V (I)	Change in production of trained and un-trained words as well as in parent-reported vocabulary	Visual analysis

*Note: **M** = methodological studies, **L** = longitudinal studies, **I** = intervention study*

5.5 ETHICAL CONSIDERATIONS

All studies were ethically approved by the Regional Ethics Board in Stockholm (study I: dnr 2012/2213-31 & 2013/1989-32; study II dnr 2012-46-31/2, 2012/2:2, 2014/1162-31/1, 2015/1401-31, & 2016/267-32; study III and IV: 2017/206-31; study V: dnr 2016/2023-31/2 & 2018/1270-32). The legal guardians of all participants gave written consent to participate and were informed that they could withdraw this consent at any time, without consequences (including any consequences to their child's care).

To ensure anonymity, some details on the participant's background information are left out. For example, the rare genetic syndromes in the participant group are not specified, as the inclusion of this information may make it possible to identify the participants. Furthermore, data on gestational age and results of brain imaging are presented on a group level instead of for each individual participant.

6 RESULTS

The results will be presented per research aim (see page 23).

6.1 VALIDATION OF THE CBR MEASURE

CBR^{UTTER} was strongly correlated to both CBR^{syl} ($r = 0.948$, $p < 0.0001$) and CBR^{utt} ($r = 0.876$, $p < 0.0001$), but the agreement was considerably lower for CBR^{utt} (ICC = 0.475, $p < 0.001$) than for CBR^{syl} (ICC = 0.936, $p < 0.0001$). Thus, the novel measure CBR^{UTTER} seems to be valid compared to CBR^{syl}, when using the measures on the same data.

The validity of CBR^{UTTER} was futher strengthened by a strong association with CB status based on babbling observation in 10-month-old children (AUC = 0.87, 95% CI 0.76–0.97). Based on positive likelihood ratios >3 and prioritizing high sensitivity, a CBR criterion of 0.14 was suggested instead of the traditionally used 0.15. The application of this 0.14 criterion in our data resulted in a sensitivity of 0.96 and a specificity of 0.70. In the predictive comparisons, not being in the CB stage at 10 months (using a 0.14 criterion) predicted speech/language difficulties at 30–36 months of age with a sensitivity of 0.71 and a specificity of 0.52.

6.2 BABBLING IN ND AND ITS ASSOCIATION TO LATER SPEECH PRODUCTION

Babbling data was compiled from the children with neurological disabilities at T1 and can be found in table 9, together with a brief summary of speech and language data at T2 and T3. Compared to data from age-matched, typically developing children, a higher proportion of children with ND had not reached babbling milestones. Five out of 18 children were not in the CB stage (as defined by $CBR \geq 0.15$), 6/18 did not use dental plosives and 4/18 did not use any plosives. All typically developing children had reached all babbling milestones. According to the Fisher's exact test, differences between the groups were significant when it comes to being in the CB stage and using dental plosives. Considering only children with DS, a slightly different pattern appeared: all children with DS were in the CB stage and 5/6 used plosives and dental plosives. The children with ND used fewer different consonants compared to the typically developing children (median 6, range 1–11, as compared to median 9, range 6–13). This pattern was the same for children with DS (median 6, range 4–7).

As can be seen in table 9, there were no clear-cut associations between CBR and later speech or language disorders. Children with both typical and non-typical speech and language at T2 and T3 could be found in both the lower and the higher CBR range at T1. Thus, higher CBR was not associated with better speech and language at age 5 or 7. However, the participants with the lowest CBR at T1 all had severe difficulties at T2/T3. On the other hand, there were also children who were non-speaking at age 5 and 7 (shown in the table as PCC = 0) who had high CBR at T1.

Table 9. Overview of babbling, speech and language data for the 18 participants with ND from three time points (T1, T2, and T3). Participants are arranged by canonical babbling ratio at T1.

	T1						T2		T3	
Participant	**Age (months)**	**CBR^{UTTER}**	**Plosives**	**Dental plosives**	**N of different consonants**	**N of different true consonants**	**PCC**	**Language difficulties**	**PCC**	**Language difficulties**
O1	14	0.00	No	No	1	1	0	Yes	0	Yes
CP5	14	0.05	No	No	2	2	0	Yes	0	Yes
CP6	16	0.06	No	No	1	1	0	Yes	-	-
O2	13	0.13	Yes	No	4	4	-	Yes	-	-
CP3	18	0.14	Yes	Yes	7	6	93	Yes	100	No
CP1	19	0.21	Yes	Yes	7	6	57	Yes	10	Yes
DS1	12	0.24	Yes	Yes	6	3	25	Yes	50	Yes
DS3	22	0.24	Yes	Yes	6	4	29	Yes	45	Yes
CD3	14	0.25	Yes	No	5	4	0	Yes	0	Yes

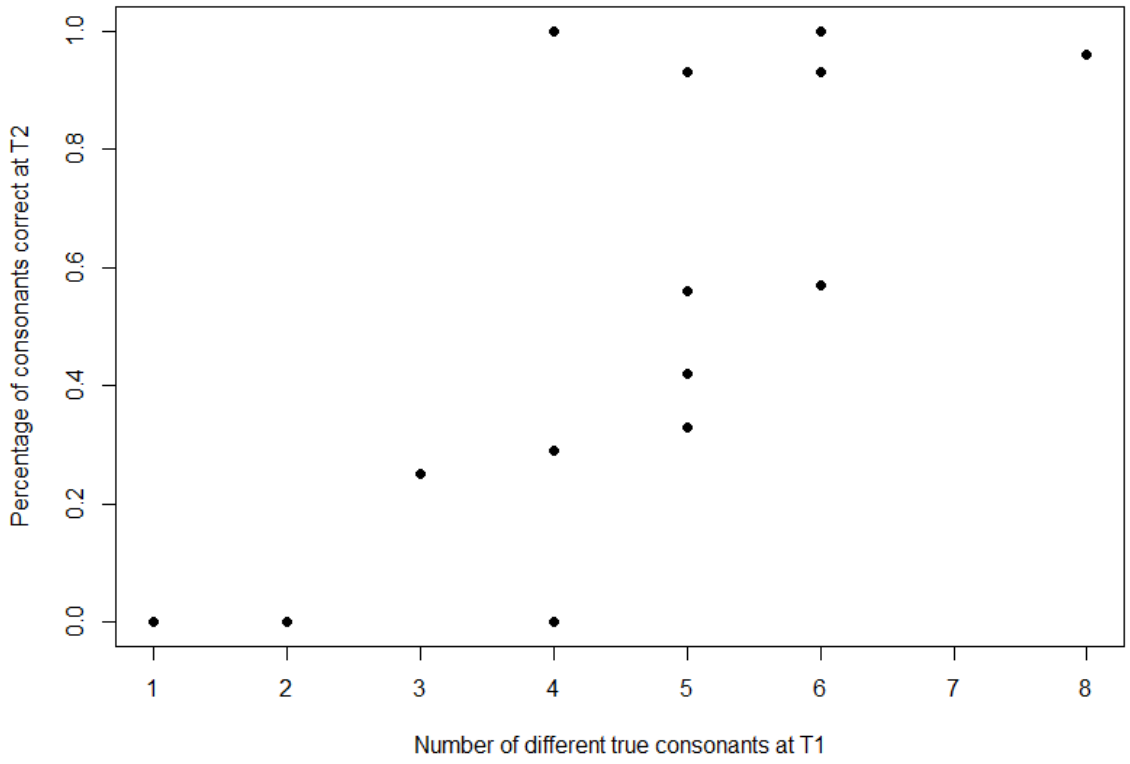
	T1						T2		T3	
Participant	**Age (months)**	**CBR^{UTTER}**	**Plosives**	**Dental plosives ***	**N of different consonants**	**N of different true consonants**	**PCC**	**Language difficulties**	**PCC**	**Language difficulties**
DS6	22	0.33	No	No	4	2	0	Yes	0.15	Yes
CD1	12	0.42	Yes	Yes	4	4	100	Yes	93	Yes
CP4	12	0.47	Yes	Yes	5	5	93	No	100	No
DS5	21	0.55	Yes	Yes	6	5	56	Yes	52	Yes
DS4	20	0.57	Yes	Yes	6	5	42	Yes	-	-
O3	16	0.64	Yes	Yes	9	7	-	-	-	-
CD2	13	0.65	Yes	Yes	9	6	100	No	100	Yes
CP2	22	0.68	Yes	Yes	11	8	96	Yes	100	Yes
DS2	21	0.70	Yes	Yes	7	5	33	Yes	67	Yes

*Note: **CBR** = canonical babbling ratio, **PCC** = percentage of consonant correct.*

Dental plosives include /t/ and /d/. Measures not completed (for example due to attrition) is marked by a hyphen.

The association between babbled consonant production at T1 and consonant production at T2 is illustrated in figure 2. This association was tested statistically for the sub-group of children with Down syndrome who were speaking at age 5 (n = 5). For speaking children with DS, there was an association between the number of different true consonants at T1 and PCC at T2 (r_s = 0.894, p = .041). Thus, children with DS who used more consonants at 12–22 months of age, also had a higher PCC at age 5.

Figure 2. Association between the number of different true consonants at 12–22 months (T1) and percentage of consonants correct at 5 years (T2).



6.3 SPEECH AND LANGUAGE DEVELOPMENT AND DISORDER IN NEUROLOGICAL DISABILITY

6.3.1 Results on speech and language tests

The results on the speech and language tests at age 5 and age 7, as well as ratings of communication ability at age 7, can be found in table 10. One participant (CP4) had results at age level for all speech and language measures at both 5 and 7 years of age. Two participants had results at age level on all tests at either 5 or 7 years of age (CP3 and CD2). Two participants had a language disorder, without co-occuring speech disorder (CP2 and CD1). This result was

stable across both the 5- and 7-year assessments. Thirteen participants had severe difficulties with both speech and language. Most participants improved their speech and language abilities between 5 and 7 years of age, but only one participant moved from non-age-appropriate results to age-appropriate results. In addition, one participant moved from an age-appropriate to a non age-appropriate result. At 7 years of age, the median rating of communication activity was 2.5 on the TOM scale. This mean that a majority had not reached stage 3 (consistent communication outside of the immediate context) and thus ratings show a high occurrence of significant communication activity limitations.

6.3.2 Received SLP services

Parents to 12/14 participants at age 7 reported that they received SLP services "once or a few times per year" or less. The median number of recorded SLP services during the last year was 5.5. Parents indicated that their children had received a wide variety of services since birth, including language assessment (9/14 participants), counselling on language/communication (9/14), AAC services (7/14), speech/language/communication training by SLP (6/14), parental education (6/14) and feeding or oral motor services (6/14).

Table 10. The participants' (n = 17) results on speech and language tests at ages 5 and 7. Participants are ordered by rated communication ability at age 7. Bold figures represent results within age norms.

		Age 7			Age 5		
TOM-level[a] at age 7	Participant	**TROG[b]**	**Sentence recall[c]**	**SVANTE[d], PCC**	**TROG-2**	**Sentence recall[c]**	**SVANTE[d], PCC**
1: *Limited functional communication.*	CD3	0	-	0	0	-	0
2:*Communicates basic needs and information to informed/familiar communication partners.*	CP5	0	-	0	0	-	0
	DS1	0	-	25	0	-	50
	DS5	0	-	56	0	-	52
	DS6	0	-	0	0	-	15
	O1	0	-	0	0	-	0
2.5: *Abilities slightly above level 2, but not at level 3*	DS2	0	-	33	0	-	67
	DS3	0	-	29	0	-	45

		Age 7			Age 5		
TOM-level at age 7	Participant	**TROG**	**Sentence recall**	**SVANTE, PCC**	**TROG-2**	**Sentence recall**	**SVANTE, PCC**
3: *Consistent level of communication relating to subjects outside the immediate context.*	CP1	0	-	57	2	-	10
	CD1	0	-	**100**	3	-	**93**
5: *Able to communicate with anyone in any circumstance using broad range of communication modes.*	CP2	0	1	**96**	4	5	**97**
	CD2	**5**	**7**	**100**	10	**8**	**100**
	CP3	**11**	4	**93**	**18**	**8**	**100**
	CP4	**13**	**10**	**93**	**16**	**12**	**100**
Not classified	CP6	-	-	-	0	-	0
	DS4	-	-	-	0	-	42
	O2	-	-	-	0	-	-

Note: Assessments not completed is marked by a hyphen. Participant O3 did not participate at 5 or 7 years of age and is therefore not included in the table. ***a*** *Therapy Outcome Measures (Enderby, 2014).* ***b*** *Test for Reception of Grammar-2 (Bishop & Garsell, 2009). Reported as number of correct blocks.* ***c****. Subtest from the Clinical Evaluation of Language Fundamentals-4 (Semel et al., 2003). Reported as standard scores (mean: 10, SD: 3).* ***d****. Swedish Articulation and Nasality Test (Lohmander et al., 2015). Reported as percentage consonants correct.*

6.4 EVALUATION OF INTERVENTION FOR CHILDREN WITH CP

A summary of the four participants' expressive vocabulary results after the parent-implemented intervention can be found in table 11. Benjamin showed a clear gain in trained words only, as well as a large increase in parent-reported vocabulary. Adam showed a clear gain in target words, but also a gain in control words. He also had a large increase in parent-reported vocabulary. Charlie showed no gains in trained or control words during the intervention. His parent-reported vocabulary showed moderate gains. David showed some gains in trained words only, but the results were uncertain due to a fluctuating baseline. His parent-reported vocabulary showed moderate gains.

Table 11. Results for the four participants regarding target words, control words and parent-reported vocabulary .

		Adam	Benjamin	Charlie	David
Target words	Number of words produced at last probe	7/10	6/8	1/9	4/8
	Improvement	Yes	Yes	No	Unclear
Control words	Number of words produced at last probe	4/10	1/9	1/10	0/8
	Improvement	Yes	No	No	No
Parent-reported vocabulary[a]	Increase from study intake to follow-up (number of words)	268	264	82	81
	Improvement	Large	Large	Moderate	Moderate

*Note: **a**. As measured by the Swedish Early Communicative Development Inventory: words & gestures (Eriksson & Berglund, 1999).*

7 DISCUSSION

7.1 DISCUSSION OF THE RESULTS

7.1.1 Validation of the CBR measure and the 0.15 criterion

The aim of the methodological studies was to validate the CBR measure. Although commonly used in clinical babbling research, CBR has seldom been evaluated. Study II showed that CBR^{UTTER} is a valid measure, when compared to CB observation. Thus, infants judged as being in the canonical babbling stage by a trained observer are very likely to also have a high CBR. CBR has rarely been compared to other babbling measures, but Lohmander and colleagues (2017a) found a similar high agreement between CBR and CB observation. Study I compared CBR^{UTTER} to two other versions of the CBR. The high agreement between CBR^{UTTER} and CBR^{syl} indicates that they may be used interchangeably. The fact that CBR^{syl} requires that all syllables in a child's production be counted, whereas CBR^{UTTER} does not, means that time and effort may be saved by using CBR^{UTTER}. CBR^{UTTER} was correlated also to CBR^{utt}, but the agreement was lower. This is not surprising, as CBR^{utt} differs from the other two measures by not having the same unit in the numerator and denominator (the number of canonical syllables divided by the total number of utterances). This means that CBR^{utt}, in contrast to CBR^{syl} and CBR^{UTTER}, may be greater than 1, and indeed often is in infants producing many reduplicated canonical utterances.

Study II is, according to a literature search, the first study that explicitly aimed to examine the validity of the 0.15 CBR criterion after it was introduced by Lynch and colleagues (1995). This is somewhat surprising, given how well-used 0.15 is as a cut-off for categorizing children as typical or atypical babblers. In study II, 0.14 was found to render slightly better specificity than 0.15, both compared to concurrent CB observation and as a predictor of speech/language difficulties at 30–36 months. For sure, the difference between 0.14 and 0.15 is small, and 0.14 as preferable over 0.15 may not hold in studies using other data. Study II does, however, show that the choice of CBR criterion matters, as it impacts outcomes. For example, the low specificity of the 0.15 criterion in study II indicates that quite a few children would be classified as being non-canonical based on CBR, although a trained observer would classify them as canonical from an overall impression. Lowering the criterion would result in higher agreement for children classified as canonical by the trained observer (higher specificity), but lower agreement for the children classified as non-canonical by the trained observer (lower sensitivity). Thus, CBR criterion levels could be problematized more in research on infant vocalizations.

Another issue worth considering in light of the results of the methodological studies is the use of CBR to create a dichotomous classification. CBR was created as a continuous measure, to describe the development of canonical babbling over time, and although a criterion for the CB stage is mentioned in studies by Oller and colleagues (Lynch et al., 1995; Oller et al., 1994), the focus was not primarily on classifying infants as canonical or non-canonical at a given point in time. Furthermore, there are other methods for determining whether a child has entered the

CB stage that are valid and easier to perform than CBR calculations, namely observation (Lohmander et al., 2017a) and parent report (Oller et al., 1998). Dichotomizing a continuous measure results in quite a lot of data loss, and the question is if a dichotomous classification based on a CBR criterion really is superior to a classification based on CB observation or on parent report.

7.1.2 Babbling and its association to speech production in neurological disability

Study I and III aimed, among other things, to describe babbling in children with ND, and its association to later speech production. Results showed that babbling was delayed in the group of children with ND, confirming previous research on children with CP (Levin, 1999; Ward et al., 2022), Rett syndrome (Bartl-Pokorny et al., 2022; Lang et al., 2019; Marschik et al., 2012), William's syndrome (Masataka, 2001) and Fragile X syndrome (Belardi et al., 2017). When it comes to Down syndrome, the results showed clear delays compared to controls on consonant production only, which is in accordance with previous research showing no or only small delays in the onset of canonical babbling in DS (CoboLewis et al., 1996; Kent & Vorperian, 2013; Lynch et al., 1995; Smith & Stoel-Gammon, 1996).

Although the participants exhibited delayed babbling milestones compared to typically developing children, some limitations in the data prevent firm longitudinal conclusions on babbling as a predictor in this group – above all the wide age span of participants at time point 1. However, some exploratory findings will be discussed. No clear associations could be seen between CBR at T1 and speech or language at T2 and T3. In particular, great variability was seen among participants who were in the CB stage at T1 – from participants with typical speech and language to participants without speech and severe difficulties with language comprehension. At T1, four participants had a CBR < 0.14. Out of these, one had an undetected severe hearing loss at the time (participant O2, CBR 0.13). The other three, who all had a CBR < 0.07, were non-speaking at T2. Thus, participants with very low CBR also ended up with severe speech and language disorder. The data from the longitudinal studies thus support a "necessary but not sufficient" view of canonical babbling as a precursor to speech in children with ND (Lang et al., 2019; Oller & Seibert, 1988).

In addition to delays in CB onset, the participants with neurological disability also to a larger extent failed to meet milestones related to consonant production (use of plosives and dental plosives). Furthermore, they used fewer different consonants. The latter variable was the only one differentiating the children with Down syndrome from typically developing controls. A similar variable (number of different true consonants at T1) was correlated to speech accuracy (as measured by the percentage of consonants correct) at T2 for the same group of children. Consonant variables have been less examined than CB in research on clinical risk groups, but there are indications that they may be a predictor of at least later consonant production. The number of different consonants (or true consonants) at an early age has been shown to be related to parent-reported expressive vocabulary at 24 months (Persson et al., 2021b, in children with hearing loss and controls with normal hearing), vocabulary at 18 months (D'Odorico et al.,

2011, in children born prematurely) and percentage consonants correct at 3 years (Lohmander & Persson, 2008, in children with cleft palate).

The research on prediction of speech and language abilities presented and discussed in this thesis has focused as babbling as a sole predictor or indicator of speech and language disorder. This is, of course, a simplification. Other variables established as predictors of child language development include for example parental responsiveness and parental speech style (Ramírez-Esparza et al., 2014; Tamis-LeMonda et al., 2001). Especially for children with ND, who often have difficulties with both language comprehension and using language as communication, more aspects need to be taken into consideration when predicting future speech and language abilities. One example of this is the research of Yoder, Warren and colleagues, examining the longitudinal development of young children with developmental delay enrolled in early intervention services (a group who considerably overlaps with the definition of ND used in this thesis). In this project, different babbling variables are analyzed as predictors, together with other language and communication variables – such as the communicative use of vocalizations, parental responsiveness, and early receptive and expressive vocabulary – controlling for factors such as maternal education and child mental age (McCathren et al., 1999; Yoder & Warren, 2004; Yoder et al., 1998). In future research on babbling as a precursor for children with ND, similar designs would be beneficial.

7.1.3 Speech and language development and disorder in children with neurological disabilities

The aims of study III and IV included describing speech and language development in a group of children with ND, enrolled within the Swedish habilitation services.

Earlier research has shown a high prevalence of speech and language disorder in ND, and the results of the longitudinal study confirm this in participants recruited in a Swedish habilitation context. In fact, out of the total of 17 children examined at either 5 or 7 years of age, only one had typical performance on all measures and at both ages. Although the high prevalence was not unexpected per se, there are indications that this study group, not selected on language or cognitive ability, may have more severe speech and language disorder than in previous studies. For example, the results from participants with DS may be compared to the participants from Næss and colleagues (2015), who were of the same age. They had a mean raw score of 12.74 (SD 9.5) on the Norwegian version of the TROG, whereas all participants in the longitudinal studies in the present project had a raw score of 2 or less on the Swedish version. Speech and language disorder were not only present as measured with speech and language tests but also on ratings of everyday communication (based on a parent interview). Although some participants with speech and/or language disorder were rated at the highest level of communication ability, 11/15 participants were rated as having communication difficulties at age 7.

Speech and language abilities did in most cases improve between 5 and 7 years of age, but the test results rarely changed from below age-level to age-level, or vice versa. The fact that

children with severe speech/language disorder do not catch up on their peers is not surprising. The faster rate of development in typically developing children compared to children with ND has for example been shown by Næss and colleagues for Down syndrome (Næss et al., 2021). This increasing gap with age could also be the reason participant CD2, who had typical language results at T2, presented with language comprehension difficulties at T3 (this participant did, however, have another genetic syndrome than DS). More interesting is perhaps the participant CP3, with improved language ability relative to peers between T2 and T3 (moving from non-typical results at age 5 to typical results at age 7). A similar catch up-effect has been shown earlier for children with CP in the area of language comprehension, albeit at a slightly lower age (Hustad et al., 2018).

Communication disorders in children with ND is a complex area of research, as the group is heterogenous and many different factors can affect the communication ability of the child. Speech and language abilities, which are in focus in this thesis, are of course important, but a child's communication is also affected by other abilities (such as non-verbal cognition, fine and gross motor abilities and perception, among others), by presence of other conditions (diagnoses such as ADHD or autism as well as comorbid somatic health conditions). Perhaps even more importantly, communication disorders are affected by the child's communicative environment (including communication partners and the different communicative contexts that they participate in).

Even when only considering speech and language ability, there is still a substantial complexity to deal with. This complexity might be what is reflected in the wide variety of measures used in research on speech, language and communication characteristics in ND. Speech and language disorder has been defined in different ways across studies and speech and language have been examined using different degrees of accuracy.

People with ND often have both speech and language disorder. From a clinical perspective, it is important to differentiate between speech and language disorder, as the type of disorder determines the focus of intervention. This differentiation is however not always straight-forward, especially not in young children.

7.1.4 Provision of SLP services to children with neurological disabilities

One of the aims of study IV was to explore SLP intervention in the group of children with ND. Although the speech and language disorders present among the participants seem to have resulted in severe communication activity limitations (as indicated in ratings of communication abilities), the frequency of SLP services was overall low. In international studies, the most common type frequency of service delivery seems to be weekly or bi-weekly sessions (see Meyer et al., 2017, studying service delivery to children with DS in Australia and Majnemer et al., 2014, studying service delivery to children and adolescents with CP in Canada). There are, however, also previous studies which report results in line with those in this thesis. In a survey of people with DS and their families in Ireland, Frizelle and colleagues (2021), 89% of respondents reported on six SLP sessions per year, or less. The results are also in line with that

of Tegler and colleagues (2018), who investigated the amount of instruction provided by SLPs in the habilitation services to children who had been prescribed communication aids. Half of the respondents had provided only one instruction session during the last year.

Parents reported a wide variety of types of services, many of which were aimed at communication partners and the child's everyday activities. A combination of direct intervention and indirect services (aiming at improving knowledge and skills in parents and other important people in the child's everyday life, to enable them to support the child's communication and participation) is in line with Swedish treatment recommendations (Eberhart et al., 2011), and has also been suggested in international research (Hustad & Miles, 2010; Majnemer et al., 2014). However, one could question whether these combined interventions are meaningful with such low treatment intensity. Studied interventions for speech sound disorder often report a high frequency of treatment, resulting in a number of sessions far exceeding what has been reported in this thesis (see for example Kaipa & Peterson, 2016). Although treatment intensity rarely have been examined for people with neurological disabilities, it is highly unlikely that children with ND would need less treatment intensity than children without ND for an intervention to be effective. When it comes to interventions aimed at improving communication skills of parents or introducing AAC, high treatment intensity has not been as explicitly advocated as in interventions for speech sound disorder, but interventions are nevertheless often reported to include more sessions than 5.5, which was the mean number of registered sessions in study IV. For example, the *It takes two to talk* program by the Hanen centre (Pepper et al., 2004) includes six to eight group sessions and three individual sessions and the Swedish *ComAlong* parent program (Jonsson et al., 2011) includes eight group sessions.

The reason for the relatively low number of reported and registered SLP sessions was not examined in this thesis, which precludes firm conclusions. Earlier research have however shown that a low frequency of SLP sessions may be caused by factors such as inadequate clinician time, large caseloads and staff shortage (Meyer et al., 2017; Ruggero et al., 2012). A possible factor in the Swedish context is the fact that few SLPs are employed by schools. When reviewing the results on SLP service delivery in the habilitation context it is important to remember that the services are multi-professional and centered around the child's needs rather than specific professions. Measuring service delivery by a specific profession will therefore not provide the whole picture of the child's services.

7.1.5 Evaluation of intervention for children with neurological disabilities

Study V aimed at evaluating a parent-implemented intervention for children with cerebral palsy and speech/language disorder. Out of the four children examined, two showed clear signs of having learned the target words and they also showed large gains in parent-reported vocabulary. One participant showed unclear evidence of having learned some target words and one participant did not show evidence of having learned the target words. These two participants did increase their parent-reported vocabulary from intervention start to follow-up, but changes were moderate. Focused stimulation has previously shown promise in children with non-

neurological disabilities such as expressive language delay and cleft palate (Girolametto et al., 1996; Ng et al., 2020; Scherer et al., 2008). There is also one previous study on children with neurological disability, namely Down syndrome (Girolametto et al., 1998). The results of the intervention study is an indication that a focused stimulation intervention may be effective for children with CP as well. The focused stimulation approach for children with CP and speech/language disorder is suitable for further studies, with a larger number of participants and more experimental control.

7.2 METHODOLOGICAL DISCUSSION

In the methodological studies, secondary data were used. This limited the selection of outcome measures for speech and language. Other, more sensitive measures, might have rendered different results. In study IV, CBR was evaluated using CB observation as the reference test. Again, other choices here might have rendered different results.

Determining which cut-off to use on a test requires balancing sensitivity and specificity. This can be done in different ways, depending on the population and condition in question. In study IV positive likelihood ratios were used to define a CBR criterion for the CB stage. CBR is most commonly used in research studies that aim to identify delayed babbling development in clinical risk groups, and therefore a focus on positive likelihood ratios and sensitivity seemed appropriate. The criterion of LR+ >3 was deemed reasonable, but of course any such cut-off is arbitrary.

When it comes to the longitudinal studies, one weakness is the small number of participants, which limits the possibility of generalizing the results. The participants in the longitudinal studies were recruited from an early age and from different habilitation centers, aiming at a representative view of children receiving habilitation support at this early age. It is, however, unclear to what extent the participants can be considered representative for the population as a whole. It was estimated that about 60 children with ND may have been eligible to participate in study I. Thus, the participants constituted approximately a third of all possible eligible participants and must thus be considered somewhat of a convenience sample. In studies of language development in children, it is often surmised that parents to children with difficulties would be more prone to participating in research than children without difficulties. Following that line of argument, the participants in the longitudinal studies would have more severe difficulties than the general population of children receiving habilitation services from an early age. On the other hand, it is not necessarily true that this applies to children who already are receiving multi-professional services, as all participants in the longitudinal studies did at the time of inclusion. Rather, it is possible that the families who chose not to participate in the study had children with more severe disabilities, as a common comment from parent declining to participate was lack of time due to many medical appointments.

Another limitation of the longitudinal studies is that children with other first languages than Swedish were excluded. The rationale for this was the lack of suitable outcome measures for speech and language for most of the common minority languages in the Stockholm region as

well as availability of SLPs with competence in these minority languages. Comparing children with Swedish as their first language to children learning Swedish as a second language without considering the latter's first language abilities would not be valid and would introduce even more uncertainty to an already heterogenous group. On the other hand, a large proportion of Swedish children are learning Swedish as a second language, and the exclusion of this part of the population is problematic in the long run.

Strengths of study III and study IV include the longitudinal perspective and the inclusion of participants regardless of cognitive level and primary condition, probably making the sample a very recognizable one for SLPs in the habilitation services. Another strength is the narrow age span for the T2 and T3 assessments. At T1, the participants were of a wider age span (12–22 months) and some caution is required when interpreting the data. This is less of a problem in study I; as all participants were older than 10 months, when CB should be established, the results are probably rather an underestimation of the true proportion of children with delayed babbling milestones. In addition, partitipants were compared to age-matched controls. In the longitudinal comparisons of babbling and speech/language this is however more problematic as it means that vocalizations from the beginning of the second year of life are (indirectly) compared to vocalizations from the end of second year of life. For this reason, no statistical analyses were done on the association between babbling and speech/language for the group as a whole.

Overall, the results of the longitudinal studies should be seen as explorative and hypothesis generating. The group of participants belongs to many different subgroups, based on medical diagnoses and based on speech/language function. For each of these subgroups, specific research questions and hypotheses could be formulated and tested with appropriate measures (given that a proper number of participants could be recruited to test the hypotheses). In the longitudinal studies, a wide variety of abilities were measured and reported, with the aim of capturing the variability within the participant group. The downside of this approach is, however, the lack of detail with which each ability was examined.

In the intervention study, one important methodological issue is the selection of outcome measures. In study V, word probes were used as the primary outcome measure. This is a rather conservative measure; a child may very well be producing a word in everyday activities but still fail to do so during the word probes. This was indeed the case for the participant Charlie, who scored low on all word probes, possibly due to a reserved personality. However, the results of the word probes were in line with the secondary outcome measure as participants who scored high on the word probes also had larger increases in parent-reported vocabulary.

Another methodological issue in the intervention study is the use of untrained words as a control behavior. The idea with control words was to provide a control for maturation. On the other hand, a child learning untrained words is a highly desirable outcome of a vocabulary intervention. The use of another type of single case research (e.g., a multiple baseline across behaviors) would perhaps have been a better option.

The intervention study was an A-B study, a design chosen for its applicability within the clinical context. Although the A-B design has limitations when it comes to drawing conclusions on causal mechanisms, its feasibility makes it a valuable tool for under-researched intervention types such as focused stimulation for children with CP.

8 CONCLUSIONS

CBR^{UTTER} is a valid measure of canonical babbling, compared to other commonly used canonical babbling measures and 0.14 seems to be a suitable criterion for the canonical babbling stage, at least if high sensitivity is prioritized.

The high occurrence of speech and language disorder in children with neurological disabilities, seen in previous literature and confirmed among the participants in the present work, entails a need for increased attention to speech and language abilities in these patients.

Children with neurological disabilities in Sweden may risk being underserved by speech-language pathologists within the habilitation services. The parent-implemented vocabulary intervention examined in this thesis is an example of an intervention with higher intensity, which can result in relevant change in patients and could be implemented within the habilitation services.

9 POINTS OF PERSPECTIVE

9.1 FURTHER RESEARCH

9.1.1 Infant vocalizations – measures and association to later speech and language abilities

Canonical babbling ratio is a commonly used measure in clinical babbling research, but it has seldom been validated. Thus, there are many unknowns when it comes to CBR methodology. In this thesis, some of these are addressed. Some unknowns remain, however, including:

- How does CBR vary with the method for determining if canonical production is present? That is, are CBR measures based on transcription or categorization comparable to those based on counting in real time?
- Does the optimal criterion of 0.14 proposed in this thesis hold true even in other data sets?
- Is the optimal criterion for determining if a child is in the CB stage the same across methods for rendering CBR?
- Is CBR a good predictor of later speech and language function in children who risk difficulties?

The association between babbling variables and later speech and language in children with ND also need to be further examined, using larger groups, enabling comparisons between different medical diagnoses and combining babbling variables with appropriate speech, language and communication co-variates.

9.1.2 Speech, language and communication in children with neurological disabilities – development and intervention

Based on the exploratory results of the longitudinal studies, the following research questions could be examined in larger (and perhaps more specific) samples:

- What type and frequency of services are provided by SLPs to Swedish children with ND and are there regional variations?
- To what extent do speech and language difficulties change during childhood in children with ND?

Another interesting further research issue from the longitudinal study includes more detailed measures of communication, both based on parent interview and parent-child interaction.

The intervention study can be replicated with more participants and with other clinicians delivering the intervention. In that way, a larger data set on the effectiveness of this intervention for children with CP could be created. Furthermore, larger studies with more experimental control could be performed to further increase the quality of the evidence. A multiple baseline design has already been mentioned, but provided a sufficient number of participants, a randomized controlled trial would provide even stronger evidence. The knowledge on how the parent-implemented focused stimulation technique works for children with CP could also be

examined by analyzing detailed data on how the parents in study V used the intervention techniques and whether this is reflected in the results of the child.

In speech and language pathology research, speech disorder and language disorder are often studied separately. While this is sometimes necessary to allow for detailed analysis, there is also a risk of missing important aspects of the speech and language ability of children who have both speech and language disorder – such as many children with ND. Another challenge is the fact that idiopathic speech and language disorders (i.e., speech and language disorders in children without significant other disabilities) have constituted separate research fields in relation to speech and language disorder in children who also have other disabilities, for example children with ND (see Shriberg and colleagues, 2019, for a discussion of this phenomenon in research on speech disorder). In the light of this challenge, the CATALISE consensus on "language disorder" as a term encompassing both children with and without other disabilities is much welcome.

9.2 IMPLICATIONS FOR CHILDREN WITH NEUROLOGICAL DISABILITIES AND THEIR FAMILIES

Research has indicated that speech and language disorder affect children's quality of life (Feeney et al., 2012), a fact that makes it important to identify and treat speech and language disorders in children with neurological disability. In this thesis, the child and family perspective was included indirectly, as the SLP ratings of communication ability was based on parent interviews. To specifically include the self-reported perspective of children with ND in future studies of speech and language characteristics and intervention would be very valuable.

The results in this thesis indicate that children with ND have a very high risk of speech and language disorder and that parents despite this report low frequencies of SLP services. Although the results of the thesis are considered exploratory, the notion that many families would wish for a higher frequency of SLP services is well in line with the experience of Swedish SLPs in the habilitation services. Furthermore, the results of study IV indicate that frequency of services is indeed in many cases much lower than recommended for interventions described in the literature. There is a need for continued examination of the service delivery for children with ND and their families to determine if this is indeed the case. Such studies should include both children with ND and their parents as informants.

9.3 CLINICAL IMPLICATIONS FOR SPEECH-LANGUAGE PATHOLOGISTS

Speech and language disorder often co-occur in children with neurological disabilities. In the longitudinal study at both 5 and 7 years of age, all participants with a speech disorder also had a language disorder, but not vice versa. Thus, if only speech production is taken into account when determining need for SLP intervention, there is a risk of missing children with significant language disorder. The presence of speech disorder is more salient to non-experts and the risk of it going unnoticed are probably smaller. However, interventions for speech disorder seems not to be delivered to children with ND to the proper extent (for a report from the Swedish

context, see Egefors, 2013). It is important that both speech and language abilities be assessed and taken into consideration in intervention for children with ND.

In the Swedish habilitation organizations in general, speech and language are rarely assessed with standardized tests or instruments. Although there are difficulties associated with using standardized tests for children with ND (see Hoffmann et al., 2020 for a discussion), there are also benefits. Above all, standardized tests help clarify the gap to typically developing peers in some children with relatively milder language difficulties. Of course, standardized tests need to be used together with measures that are sensitive to clinically relevant changes, for example after an intervention, and measures of the consequences of speech and language disorder in a child's everyday life. This notion is however not unique to children with ND. To find relevant measures for assessing speech, language and communication in children with ND remains an important issue for the further development of the field.

For many children with ND, AAC is an essential asset. The fact that AAC is not examined to a great extent in this thesis does not mean to imply otherwise. SLPs within the habilitation services in Stockholm work with a wide variety of areas, including AAC, but there seems to be a problem when it comes to providing more intensive interventions. The focused-stimulation intervention examined in this thesis might be suitable for implementation in the Swedish habilitation context. It consists of ten sessions in total, some of which could be possible to deliver in a digital format. The implementation does, however, require that it is possible for SLPs to schedule interventions of this length.

In this thesis, babbling is examined as a precursor to speech. The exact role of babbling in children with ND does however remain to be evaluated. This thesis and earlier research indicate that the presence of babbling milestones such as canonical babbling does not guarantee favorable speech and language development. However, very few canonical syllables and consonant sounds might be a sign of a less favourable development.

9.4 CLINICAL IMPLICATIONS FOR OTHER HEALTH CARE PROFESSIONALS

Children with ND often have complex difficulties. Even though this thesis is focused on speech and language in particular, the influence of other abilities and environmental factors must be stressed. This means that each child with ND requires an individualized treatment, and that different areas of intervention need to be prioritized in relation to each other. To achieve this, the multi-professional team, for example within habilitation services, is essential. However, team work is also needed between the habilitation services and other parts of health care, as people with ND often have need of many different specialists. One such example became evident during the work with this thesis, namely the need of cooperation when it comes to hearing status. For some participants with ND, hearing had not been examined since infancy, although the participants had speech or language disorder. It is also known from clinical experience, that hearing status is not always top of mind during habilitations interventions. Thus, more cooperation between hearing services and other habilitation services would be beneficial.

9.5 IMPLICATIONS FOR HEALTH CARE PLANNERS

In the habilitations services in Stockholm today, speech and language are not routinely assessed in children with ND. One exception is the CPUP quality register for children with CP, where SLPs recently have begun reporting ratings on functional communication and speech production. With the help of the CPUP register, prevalence of communication difficulties in Swedish children with CP has been estimated (Kristoffersson et al., 2020) and the prevalence of rated speech disorder may be estimated in the future.

The lack of routine evaluations for most children with ND does, however, imply that the exact prevalence of speech and language disorder in children receiving habilitation services in Sweden is unknown – although as has been shown in this thesis it is probably very large. There may thus be many children with ND who have undetected or unrecognized speech and language disorder. In addition, this thesis has shown that children with neurological disability and identified speech and language disorder risk being underserved when it comes to treatment for these disorders.

In Sweden, SLP services are roughly devided into two areas. Children with speech and language disorder without additional neurodevelopmental disability receive services from SLPs employed in primary care or at hospitals, whereas children with diagnoses such as intellectual disability, cerebral palsy or autism receive habilitation services (that often include SLP services). Often, these two types of services are mutually exclusive. Today, however, researchers agree that language disorder is rarely isolated and that the groups of children with language disorder and children with disabilities overlap considerably (Bishop et al., 2017; Bishop et al., 2016).

In addition, as SLP services within habilitation are organizationally separate from other SLP services, the two may differ when it comes to allocated resources and SLP availability. This means that the actual services a child receives may be highly dependent on whether they have an isolated language disorder or a language disorder plus an additional diagnosis. Traditionally, habilitation services have been more focused on indirect interventions aimed at the activity and participation levels and less on direct intervention (such as speech and language training performed by an SLP). There is, however, no evidence suggesting that children with speech/language disorder and ND would not benefit from direct SLP intervention. Similarly, there is quite a lot of evidence implying that services focusing on alleviating the consequences of speech and language disorder is important for children with developmental language disorder as well (see for example Law et al., 2019; Pickstone et al., 2009). Ideally, both these types of services would be available to children with speech and language disorder based on need, and not based on the presence of additional diagnoses.

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