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PhD dissertation

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Parent-implemented focused stimulation in toddlers with cleft palate

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ABSTRACT

This dissertation presents three studies concerned with early intervention in toddlers with cleft palate ± cleft lip (CP) and delayed speech and/or language acquisition. The central aim of the PhD project was to assess both short- and long-term effects of a parent-implemented focused stimulation (FS) program in which productive vocabulary and consonant inventory were targeted directly. The intervention group were compared to a control group of toddlers with CP who were matched in pairs on language development but did not receive direct intervention. The second aim was to investigate if reliable and valid early identification of intervention need was possible in toddlers with CP.

Fifty-five toddlers with CP in total were included in this project. Speech-language performance was evaluated at pre-test, immediately after intervention (four months after pre-test), and six months after intervention (ten months after pre-test). Fourteen toddlers received intervention (INT group), and fourteen toddlers with CP constituted the control group (CONT group). Twenty-two toddlers participated in the validation of a screening procedure to identify intervention need based on speech-language performance.

The screening procedure was a valid and reliable tool for identifying toddlers in need of intervention. Immediately after intervention, the INT group had significantly higher gain scores with large effect sizes for two outcome measures compared to the CONT group: lexical age, a measure of observed productive vocabulary, and true consonant types in words. Odds of need for intervention were eight times lower in the INT group than in the CONT group. Six months after intervention, measures of speech accuracy, reported productive vocabulary, and receptive language were not significantly different between the INT group and the CONT group. Toddlers with CP had significantly poorer speech accuracy than toddlers without CP, and only a few toddlers with CP performed within two standard deviations of the mean of toddlers without CP.

In conclusion, early identification of toddlers with CP who need intervention was possible, but the predictive value of the screening tool is still unknown. Parent-implemented FS had short-term effect on the speech-language performance of toddlers with CP, but no long-term effects were found. Possible explanations for the lack of long-term effects are discussed, but the results of this project suggest that early intervention should not be ventured without careful consideration of intervention method, dose frequency and total intervention duration, and intervention targets.

RESUME

Denne afhandling indeholder tre studier der omhandler tidlig intervention hos småbørn med ganespalte ± læbespalte (GS) og forsinket tale- og/eller sprogudvikling. Det centrale formål med ph.d.-projektet var at måle kort- og langtidseffekt af forældreimplementeret focused stimulation (FS) der var direkte målrettet både produktivt ordforråd og konsonantinventar. Interventionsgruppen blev parvist matchet med en kontrolgruppe af småbørn med GS baseret på deres sproglige udvikling. Kontrolgruppen modtog ingen direkte intervention. Et andet formål med afhandlingen var at undersøge om det var muligt at foretage valid og reliabel identifikation af hvilke småbørn med GS der havde brug for intervention.

I alt 55 småbørn med GS deltog i projektet. Tale- og sprogfærdigheder blev undersøgt ved prætest, umiddelbart efter interventionen (fire måneder efter prætest) og 6 måneder efter interventionens afslutning (10 måneder efter prætest). 14 småbørn med GS modtog intervention, mens 14 småbørn med GS udgjorde kontrolgruppen. 22 småbørn med GS deltog i valideringen af en screeningsprocedure der skulle identificere interventionsbehov ud fra tale- og sprogfærdigheder.

Screeningsproceduren syntes at være et validt og reliabelt værktøj til identifikation af interventionsbehov hos småbørn med GS. Umiddelbart efter interventionen havde interventionsgruppen statistisk signifikant højere tilvækstscorer med stor effektstørrelse for to mål sammenlignet med kontrolgruppen: Ordforrådsalder, et mål for observeret ordforråd, og antal forskellige "true" konsonanter i ord sammenlignet med kontrolgruppen. Chancen for ikke at have behov for intervention efter fire måneder var otte gange større i interventionsgruppen end i kontrolgruppen. Seks måneder efter interventionens afslutning fandtes ingen statistisk signifikante forskelle mellem grupperne på mål for udtalekorrekthed, rapporteret produktivt ordforråd, sprogforståelse eller behov for yderligere intervention. Småbørn med GS havde statistisk signifikant lavere udtalekorrekthed end småbørn uden GS på samme alder, og kun få småbørn med GS lå inden for to standardafvigelser af den gennemsnitlige udtalekorrekthed for småbørn uden GS.

Samlet set peger resultaterne af dette projekt på at behov for intervention hos småbørn med GS kan identificeres tidligt, men prædiktionsværdien over tid er stadig ikke kendt. Forældre-implementeret FS viste kortvarige effekter, men ingen langtidseffekter. Mulige forklaringer på de manglende langtidseffekter af interventionsprogrammet diskuteres i afhandlingen, men afhandlingens resultater peger samtidig på at tidlig intervention ikke bør iværksættes uden grundige overvejelser af interventionsmetode, interventionsfrekvens og -varighed samt interventionsmål.

LIST OF STUDIES

1. Jørgensen, L. D., & Willadsen, E. (2017). Development and validation of a screening procedure to identify speech-language delay in toddlers with cleft palate. *Clinical Linguistics & Phonetics, 31*(10), 743-760, doi: 10.1080/02699206.2017.1318174

2. Jørgensen, L. D., Scherer; N. J., & Willadsen, E. (2017). *Early intervention in toddlers with cleft palate – Short-term effects of a parent-implemented focused stimulation program*. Manuscript.

3. Jørgensen, L. D. (2017). *Early intervention in toddlers with cleft palate – Long-term effects on speech accuracy, productive vocabulary, and receptive language.* Manuscript.

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Finally, I wish to acknowledge the invaluable contributions of my predecessors in the field of early intervention in toddlers with CP. Conducting these studies is no small feat, as it is made of a special blend of hard work, heterogeneity, and humongous lumps of pure joy. I could not have completed this project without your previous works.

INTRODUCTION

The acquisition of speech and language is one of the more astonishing accomplishments of early childhood. This development progresses from the cries, hiccups, and coos in early infancy over canonical babbling at six months, the first words appearing around age one to beginning word-combinations before age two. At three years of age, most Danish children will have acquired a vocabulary of 500-700 words (Bleses et al., 2008). They speak in sentences and have largely acquired the Danish phonological system (Clausen & Fox-Boyer, 2017).

It is important for toddlers to possess good communications skills for several reasons. First of all, good communication skills enable toddlers to convey their basic needs to their surroundings which may be of particular importance in Denmark where more than 90% of all 2-3 year-olds attends pre-school[1]. Good communication skills also contribute to further speech-language development. Intelligible child utterances elicit language-facilitating responses from caregivers (Warren & Brady, 2001; Frey, Kaiser, & Scherer, in press). Thus, possessing good communication skills leads to even better speech-language development. Conversely, poor communication skills lead to poorer speech-language development.

About 5-7% of pre-schoolers exhibit speech-language delay (Law, Boyle, Harris, Harkness, & Nye, 2000). Delays in speech-language acquisition are associated with later speech-language difficulties and reading difficulties, and affect peer relations, education level, and choice of vocation (Rescorla, 2011; Laws, Bates, Feuerstein, Mason-Apps, & White, 2012; Conti-Ramsden & Durkin, 2012). Known risk factors are hearing loss, cognitive impairment, autism spectrum disorders, neurological disorders, and physical malformations, including craniofacial disorder such as cleft palate ± cleft lip (Rescorla, 2011).

The body of this dissertation is made up of three studies concerned with identification and treatment of speech-language delay in toddlers with cleft palate ± cleft lip. The first study reports on the development and validation of a speech-language screening procedure for identifying need for intervention in toddlers with cleft palate ± cleft lip. The second study investigates the short-term effects of a parent-implemented early intervention program on reported and observed productive vocabulary, consonant inventory, and continued need for intervention. The third study investigates the long-term effects of the intervention program on speech accuracy, reported productive vocabulary, receptive language, and continued need for intervention. The first part of this introduction describes the background as well as the theoretical and empirical frame for these studies. The second part provides an overview of the included studies as well as a description of participants in this project and their distribution

[1] Source: Statistics Denmark

across groups and studies. Finally, results from studies are presented and discussed in terms of relevance for current practice and future research.

BACKGROUND

CLEFT PALATE ± CLEFT LIP

Cleft lip and/or cleft palate is one of the most common congenital malformations. In Denmark, about 2/1.000 babies are born with cleft lip and/or cleft palate (Jensen, Kreiborg, Dahl, & Fogh-Andersen) which corresponds to about 121 babies each year in a birth cohort of 60.500 new-borns[2]. About one third of these babies are born with cleft lip only, one third are born with cleft lip in combination with cleft palate, and one third are born with cleft palate only. More boys are born with combined cleft lip and palate whereas more girls are born with cleft palate only. As speech is not typically affected in children with cleft lip only, the term cleft palate (CP) will be used henceforth in this dissertation to refer to cleft palate ± cleft lip.

The palate develops between the fourth and twelfth week of pregnancy. The primary palate fuses with the two palatine processes in a Y-shaped pattern (and upwards with the nasal septum) to form the roof of the mouth and separate the nasal and oral cavities. Cleft palate occurs when this fusion is incomplete and can vary from a cleft uvula to a complete bilateral cleft along the entire Y-shape (Mossey, Little, Dunger, Dixon, & Shaw, 2009; Atkinson & Howard, 2011) (see figure 1).

Figure 1. The palate with Y-shaped fusion lines seen from below.

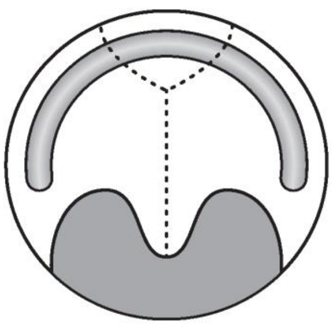

Illustration by Jakob Rønlov.

Feeding, middle-ear function, hearing, babbling and speech development, dental and jaw development, as well as facial appearance in babies with cleft lip, are likely to be affected in babies with CP. The cleft is surgically closed to provide babies with optimal potential of development of these functions and structures, including a more normal facial appearance. While it is commonly recognised that speech-language development would benefit from

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[2] Mean of live birth cohorts from 2007-2016 (Source: Statistics Denmark).

surgery performed before the age where babies typically begin to produce canonical babbling (see below), evidence for optimal timing of palatal surgery is still virtually non-existent (Leow & Lo, 2008; Lohmander, 2011; Peterson-Falzone, 2013). Thus, surgical timing varies considerably between countries and centres. In addition to surgery, children with CP will often need aggressive monitoring and treatment of otitis media with effusion, speech therapy, and orthodontic management. Secondary surgery for improving speech or as part of orthodontic treatment may further be necessary. The ultimate goal of treatment is to ensure that the cleft condition does not prevent a child born with a cleft from living a “normal” life (Shaw, Semb, & Lohmander, 2016).

TREATMENT OF CLEFT PALATE ± CLEFT LIP IN DENMARK

In Denmark, treatment for cleft lip and/or cleft palate is centralised. Health care professionals are required by law to refer all babies born with a cleft to one of two follow-up centres: Aarhus in the Western part of Denmark and Copenhagen in the Eastern part. All children have surgeries at Rigshospitalet, Copenhagen University Hospital. Palatal surgery is currently performed between 12-14 months according to standard protocol[3]. Specialised nurses, speech-language pathologists (SLPs), orthodontists, ear-nose-throat specialists, and psychologists see the children at the follow-up centres at regular intervals. Although surgery and orthodontic treatment are centralised, speech-language therapy is not.

The specialist SLPs in the centres evaluate speech-language development and provide counselling to parents. If speech-language intervention is needed, the specialist SLP will refer the child to local SLP services. Local SLP services are free of charge and must be provided to “…children whose development requires special consideration and support” (The Danish Primary Education Act (*Dan. Folkeskoleloven*), BEK no. 999, 15/09/2014).

Early identification and intervention for speech-language difficulties have been high on the political agenda in Denmark for the last decade. This is demonstrated by screening tools developed for toddlers (The Danish version of the MacArthur-Bates Communicative Developmental Inventories (CDI): Fenson et al., 2007; Andersen, Vach, Wehberg, & Bleses, 2006) and 3-6 year-olds (www.sprogpakken.dk) as well as a large scale research project involving 12.000 0-6 year-olds aimed at developing pedagogical practices and evaluation as well as parent involvement (see www.fremtidensdagtilbud.info, *Eng. The future of daycare services*). However, few children with CP below age three receive direct intervention services despite this clinical group being at risk of speech-language delay/disorder.

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[3] In this project, some participants were also part of the TOPS project. TOPS (Timing Of Primary Surgery for cleft palate) is an international randomised controlled trial where participants are randomised to surgery at six or 12 months.

Impact of cleft palate ± cleft lip on early speech-language development

Speech

Traditionally, the description of CP speech has focused on the consequences of velopharyngeal dysfunction (VPD) on articulation. However, the development of speech begins long before the baby begins to produce meaningful utterances. It is widely accepted that early infant vocalisations are “precursors to speech”, and that these prelinguistic vocalisations develop systematically in stages (Oller, Eilers, Neal, & Schwartz, 1999). Early speech development is primarily shaped by biological constraints (Kent & Murray, 1982), before gradually being more influenced by the ambient language (Locke, 1983). Thus, the development of meaningful speech is dependent on babies’ control of phonation and articulation and phonological learning (Stoel-Gammon & Sosa, 2007). Correlations between pre-speech measures and later speech-language proficiency have been reported in numerous studies. In general, more and varied pre-speech productions lead to superior speech-language proficiency later in childhood (see Stoel-Gammon, 2011).

Babies with CP are born with an oronasal coupling which poses direct limitations on speech development. In early infancy, vocalisations are predominantly vocalic and nasal due to the shape of the vocal tract (Kent & Murray, 1982). Between four and six months of age, the larynx and velopharynx gradually separate which enables the baby to produce both oral and nasal consonants and impound intraoral air pressure to produce oral stops (Kent, 1981). The oral-nasal contrast is fundamental as it is present in almost all languages around the world (Maddieson, 2008). Further, oral stops are among the most frequently used sounds in both babbling and early words in babies without CP acquiring different languages (de Boysson-Bardies & Vihman, 1991). The contrast between sonorants and obstruents represents the least complex level of distinctive phonological features and is thus vital for producing phonological contrasts in words (Dinnsen, Chin, Elbert, & Powell, 1990). However, at six months, i.e. before palatal surgery, babies with CP cannot produce the oral-nasal contrast or impound intra-oral air pressure for stop production because of the oronasal coupling. This is reflected in the lower frequency of oral stops and the higher frequency of nasals in babbling of one-year-old babies with CP compared to babies without CP (Chapman et al., 2001, Hutters, Bau, & Brøndsted, 2001). Around six months, an abrupt shift from glottal to supra-glottal consonant-like vocalisations in babies without CP has been reported (Holmgren, Lindblom, Aurelius, Jalling, & Zetterström, 1986). In contrast, one-year-olds with CP produce more glottals than their peers without CP (Chapman et al., 2001; Hutters et al., 2001, Lohmander, Ohlsson, & Flynn, 2011; Willadsen & Albrechtsen, 2006). Although these productions seem to decrease with age, this decrease is seen at a much later age than in babies without CP (Lohmander-Agerskov, Söderpalm, Friede, Persson, & Lilja, 1994; O’Gara & Logemann, 1988; Smith & Oller, 1981). A very important milestone in pre-speech development is canonical babbling where babies produce well-formed syllables with rapid speech-like transitions between fully resonant vowels and consonant-like elements. Canonical babbling appears between age five and ten months (Oller et al., 1999). Strong correlations have been found between onset, frequency, and variation of canonical babbling and later speech-language proficiency (e.g. Stoel-Gammon, 1998a; Vihman & Greenlee, 1987). Delayed onset of canonical babbling beyond age 10 months is a risk factor for profound hearing impairment (Eilers & Oller, 1994) as well as for developmental delays, including speech-language impairment (Oller et al., 1999). Babies with CP produce less canonical syllables (Chapman et al., 2001; Scherer, Williams, & Proctor-Williams, 2008a), and they may reach the canonical babbling stage later than their peers without CP (Chapman et al., 2001), although the latter is possibly not true for babies who receive early closure of the soft palate at four months (Chapman, Willadsen, Krantz, & Hardin-Jones, 2009; Willadsen & Albrechtsen, 2006; Lohmander et al., 2011).

Hence, before they produce their first words, babies with CP as a group already show delays in speech development as a direct result of the oronasal coupling. They produce fewer oral stops, more glottals and nasals, and fewer canonical syllables than their peers without CP. In addition, they produce fewer coronals (Chapman et al., 2001; Hutters et al., 2001; Lohmander et al., 2011; Willadsen & Albrechtsen, 2006) which is likely also due to the oronasal coupling although the precise cause is still debated (see Chapman & Willadsen, 2011). Although many children with CP will obtain typical speech-language development after palatal surgery, speech delays may persist after palatal surgery because of delayed hard palate closure, fistulae, or VPD (Chapman & Willadsen, 2011). However, delays may also persist as an indirect effect of the oronasal coupling on phonological learning (Harding & Grunwell, 1998; Chapman, 1993). This means that even though speech development is initially affected by the oronasal coupling, over time, it also affects phonological learning as toddlers develop phonological rules around their articulation capacity. Thus, studies have reported lower PCC scores (Percent Consonants Correct: Shriberg & Kwiatkowski, 1982) in 2 and 3-year-olds with CP (e.g. Chapman, 1993; Chapman & Hardin; 1992; Klintö et al., 2014; Willadsen, 2012) and use of more phonological processes at age 3 compared to peers without CP (Chapman, 1993; Klintö et al., 2014).

VOCABULARY

Vocabulary development is closely linked to speech development. There are substantial overlaps between the sounds and syllable shapes produced in babbling and babies' early words (Ferguson & Farwell, 1975; Leonard, Schwartz, Morris, & Chapman, 1981; Vihman, Macken, Miller, Simmons, & Miller, 1985), and it is assumed that well-practised canonical syllables become "building blocks" for early words (Vihman, 1992). When babies' repeat the same syllable over and over again, the practice and eventual mastery of this syllable help create an essential link between articulatory movement and the resulting acoustic signal: the auditory-articulatory feedback loop (Fry, 1966; Stoel-Gammon, 1998b). In addition, well-practised syllables are also thought to guide babies' attention toward adult utterances consisting of these syllables. Thus, these building blocks act as a perceptual "articulatory filter" (see Vihman, 2017). Because of the characteristic composition and smaller amount of canonical babbling in babies with CP, their first words are likely to begin with nasals, approximants, and vowels, and more labials, velars, and glottals compared to peers without CP (Estrem & Broen, 1989; Hardin-Jones & Chapman, 2014; Willadsen, 2013), and their vocabulary development is likely to be slower (Broen, Devers, Doyle, Prouty, & Moller, 1998; Hardin-Jones & Chapman, 2014; Jocelyn, Penko, & Rode, 1996; Lu, Ma, Luo, & Fletcher, 2010; Morris, 1962; Neiman & Savage, 1997; Scherer & D'Antonio, 1995).

Vocabulary development is also associated with parent-child interaction. In particular, the amount and quality of parents' contingent responses to child utterances are associated with later vocabulary measures (see Topping, Dekhinet, & Zeedyk, 2013). Contingency or responsiveness refers to parent communication directed at the child's immediate focus: being responsive to what the child is attending to. In contrast, directive parent communication attempts to control the interaction and to redirect the child's attention. Contingent responses are likely to be expansions or recasts[4] which are positively associated with vocabulary development (Carpenter, Nagell, & Tomasello, 1998). For instance, children of parents who often recasted and expanded child vocalisations at 13 months were more likely to acquire a vocabulary of 50 words faster than children whose parents were less responsive (Tamis-LeMonda, Bornstein, & Baumwell, 2001). Similarly, higher levels of parent phonetic contingency were associated with larger increase in language development over twelve months in both typically developing children and children with Down syndrome (Velleman, Mangipudi, & Locke, 1989). Furthermore, studies have reported that the composition of babbling end early speech also affects parents' responses to child utterances. Gros-Louis, West, Goldstein, and King (2006) investigated parents' responsiveness to their 9-months-old babies. They reported that parents responded to CV-like syllables with more language-facilitating responses whereas their responses to vowel-like vocalisations were less language-facilitating. As children with CP as a group produce fewer canonical syllables than peers without CP (Chapman et al., 2001; Scherer et al., 2008a), they are likely to engage in fewer conversational interactions with parents which, in turn, may lead to fewer language-facilitating responses from parents (Rescorla & Ratner, 1996; Warren & Brady, 2001). Chapman and Hardin-Jones (1991) reported that parents of toddlers with CP were as responsive as parents of toddlers

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[4] See definitions of language support strategies in table 1, p. 16.

without CP but that they had fewer child communication attempts to respond to. Frey et al. (in press) reported that parents were highly responsive to their 17-37 months-old toddlers with and without CP, but that intelligible utterances yielded more language-facilitating responses than unintelligible utterances. Frey et al. (in press) found that toddlers with CP produced significantly fewer and less intelligible utterances than peers without CP, again demonstrating how children with CP may engage in less parent-child interactions than their peers without CP, but also, perhaps even more importantly, less language-facilitating interactions.

It is important to bear in mind that parent-child interaction is transactional in nature: while parental responsiveness is associated with increased child vocalisations, these increased vocalisations again elicit more parental responses. This process is called a transactional model of responsivity (Sameroff & Fiese, 2000; Tomasello & Farrar, 1986; Veneziano, 1988). Hence, a causal relationship cannot be assumed. It is likely that the child's communication skills to some extent influence parents' ability to engage in meaningful parent-child interactions (Fey, 1986; Hammer et al., 2001; Roberts & Kaiser, 2011).

EARLY SPEECH-LANGUAGE INTERVENTION

EARLY INTERVENTION MODELS

The observations of parent-child interactions and associated language development have informed models of early language intervention. Early intervention models can be described by their location on a "continuum of intrusiveness" (Fey, 2003). In one end, models are highly intrusive and trainer-oriented, employing direct teaching of pre-determined targets in the clinic. In the opposite end are minimally intrusive, child-oriented models with indirect teaching in a naturalistic setting. The goal is to establish language-facilitating interaction patterns and thus involves no pre-selected targets (e.g. Buschmann et al., 2008; Girolametto, 1988). Trainer-oriented approaches have largely been abandoned as an option for toddlers because not all toddlers are able to participate in such highly structured interventions, and because of low generalisation of targets into everyday contexts (Fey, 2003). On the other hand, minimally intrusive, general language stimulation has shown effect on the number of parent-child interactions, expressive vocabulary, and early grammatical constructions whether implemented by clinicians or parents (e.g. Robertson & Ellis-Weismer, 1999; Baxendale & Hesketh, 2003; Buschmann et al., 2008). However, general language stimulation may not be effective when there is a need to target specific linguistic forms (Fey, Cleave, Long, & Hughes, 1993; Finestack & Fey, 2013). In this case, hybrid intervention approaches that fall somewhere in the middle on the "continuum of intrusiveness" may be more appropriate. Focused stimulation (FS) and environmental milieu teaching (EMT) are hybrid interventions that have been used with different clinical groups (e.g. Fey et al., 1993; Girolametto, Weitzman, &

Clements-Baartman, 1998; Kaiser & Roberts, 2013; Hancock & Kaiser, 2002), including toddlers with CP (e.g. Kaiser, Scherer, Frey, & Roberts, 2017; Scherer, 1999). Table 1 provides definitions of key concepts in early intervention.

Table 1. Definitions of key concepts in early intervention.

Concept	Definition	References
Environmental arrangement	Select, arrange, and manage appropriate toys to increase the child's engagement and thereby likelihood of initiating communication.	Scherer & Louw, 2011
Responsive interaction	Encourage child utterances and follow the child's lead. Respond contingently to every child utterance with related comments, questions, or expansion/recasting.	Tannock, Girolametto, & Siegel, 1992
Prompts	Strategies to facilitate child language use, e.g. modelling or incidental teaching.	Kaiser & Hancock, 2003
Modelling	Present frequent, highly-concentrated presentations of a language target while still following the child's lead in terms of topic and activity	Girolametto, Pearce, & Weitzman, 1996
Expansion*	Repeat the child's words and basic meaning but modify the child's sentence by changing structural or semantic details without changing the sentence modality. Child: "Cat want mik" Adult: "The cat wants some milk"	Cleave, Becker, Curran, Van Horne, & Fey, 2015
Recasting*	Repeat part of or an entire child utterance while adding new syntactic, semantic, or phonological information, maintain the basic meaning expressed by the child. Child: "Cat want mik" Adult: "Does the cat want some milk?"	Fey, Krulik, Loeb, & Proctor-Williams, 1999
Speech recasting	Repeat a child utterance while adding new syntactic or phonological information. Child: "Cat want mik" Adult: "The cat wants milk"	Scherer & Kaiser, 2010; Yoder, Camarata, & Gardner, 2005
Extensions/expatiations	Respond in continuation of the child's topic and add new information but do not necessarily repeat any of the child's words. Child: "Cat want mik" Adult: "Yes, I think he is really thirsty"	Cleave et al., 2015

*The terms expansion and recasting are used inconsistently in the literature on language support strategies. In particular, expansion is used as the definition of recasting above.

FS and EMT are similar in that they are designed to be used in everyday activities to teach functional language, and they use environmental arrangement, responsive interaction, and prompting to facilitate language use (Scherer & Kaiser, 2007). In FS, the adult provides multiple models of the language target that are selected according to the child's current level of functioning while at the same time following the child's lead. Thus, FS includes some adult control compared to more naturalistic intervention approaches. EMT is also based on adult-child interactions but involves more adult control in terms of a strict prompting hierarchy. This hierarchy is comprised of all or some of the following prompting levels: 1. When the child shows interest in an object, the adult waits 30 seconds for a response from the child. 2. If the child does not respond, the adult asks “what do you want?” and waits again. 3. If the child does not respond, the adult elaborates the request by asking “what is it?” and waits again. 4. If the child still does not respond, the adult models the response and again prompts the child. When the child responds, the adult praises a correct response before handing the desired object to the child. An incorrect response evokes another model and request for imitation. The most salient difference between the models is that in FS, the child is never required to imitate the target form whereas in EMT, imitation is required (Finestack & Fey, 2013). In FS, the adult responds to the child’s incorrect attempts at the target by recasting. In EMT, the child is expected to use the target correctly during each teaching episode. If a target is not realised correct, the adult models the target and requires the child to imitate. A recast is then used to expand the child’s utterance (Finestack & Fey, 2013).

THEORIES OF LANGUAGE LEARNING BEHIND EARLY INTERVENTION

Early intervention models are based on different theoretical paradigms on language learning (Fey, 1986; Scherer & Kaiser, 2007). Three theories are mentioned here because they figure prominently in the literature, and because they demonstrate quite well how these substantially different language learning paradigms have influenced early intervention models.

The principle behind *operant learning theory* (behaviourism) as described by Winokur (1976) is that verbal behaviour subject to the same principles that govern all behaviours. Language is not viewed as different from behaviour in general. The child is believed to be a passive learner who acquires knowledge and skills automatically through the influences of the environment. The central element within operant learning is the verbal operant. The verbal operant is comprised of the causal relationship between a stimulus and a verbal response, mediated by reinforcement. The idea is that a child will never name an object without hearing someone else (a reinforcing person) name the object first, e.g. the child sees a dog (the stimulus) and says “dog” (the verbal response) must be mediated by reinforcement. Reinforcement strengthens the relationship between the stimulus and the child’s response, and thus, increases the likelihood of the response occurring again (Winokur, 1976). When used in intervention, the child should be presented with an object or picture that is simultaneously named by the clinician, and the child should be encouraged to imitate. Correct imitation should be reinforced, while incorrect imitation should be corrected or ignored (Fey, 1986). Operant learning theory entails that learning is dependent on the environment, but in an intervention context, targets should be highlighted without the distractions of a natural context (Camarata & Nelson, 1992).

Within social learning theory (Bandura, 1977), the child is viewed as an active participant in learning with the capacity for selecting, organizing, and transforming stimuli rather than just responding to them. Behaviour is learned through observation of models in a complex interaction of cognitive factors with the environment. Whether learning occurs is dependent on the child's level of attention, retention (mentally rehearsing and retaining new behaviour), motor reproduction, and motivation. Learning is not dependent on any response or reinforcement. Social learning theory is concerned with how social and self-generated factors can affect attention and motivation processes and thereby the child's ability to learn. In intervention, this means that a clinician must motivate the child to attend to the target. This can be achieved by choosing a model the child identifies with, e.g. a parent, to ensure the child's attention, and by frequent target modelling to give the child an opportunity to mentally rehearse the target. Finally, the model should, after numerous presentations of the target, probe for imitation and reinforce correct production to help enable motor reproduction, provide feedback concerning accuracy, and motivate further use of the target (Bandura, 1977; Fey, 1986).

The social-interactionist perspective emphasizes the importance of social use and context of language for development (Bloom & Lahey; 1978; Bruner, 1983). This perspective differs from operant learning theory and social learning theory in that it specifically accounts for language development and not development in general. Language is viewed as a complex system involving language content, form, and use. Language content consists of referential knowledge of word meanings, and relational knowledge of relationships between word meanings. Language form refers to the rule system (phonology, morphology, and syntax) relating sounds and sound sequences to meaning. Language use refers to pragmatics and reflects the social nature of language. Language cannot be understood without consideration of social function and influence of social context on content and form. Developmental change is believed to occur as the child actively explores the physical, social, and linguistic world, and language learning depends on the ability to understand that others have independent thoughts and motivations and the ability to extract patterns related to language forms from the context of social routines (Tomasello, 1992).

Within the social-interactionist perspective, the clinician is a facilitator more than a teacher and manipulates the environment to facilitate the child's perception of relations between language content, form, and use. Form should never be separated from meaning, and target behaviour should be modelled frequently in meaningful contexts. Language should be used to describe objects and events to which the child is attending in order to help the child induce relations between events and language form and content. Contingent verbal responses (e.g. expansions) and non-verbal consequences should consistently follow child communication, and the complexity of adult speech should reflect the child's current language level to increase salience of new form and content. Activities should be selected according to the child's interests to facilitate natural communication (Fey, 1986). Finally, parents are often involved as primary interventionists to enhance the naturalness of interactions and thus motivation for language learning (Abbeduto, Keller-Bell, Richmond, & Murphy, 2006).

It should be clear from the above that the social-interactionist perspective of language learning has massively influenced naturalistic early intervention procedures, including hybrids (Girolametto et al., 1996), although the different theories overlap to some degree. In particular, modelling fit well within all three paradigms. Operant learning theory has influenced the more intrusive intervention approaches but also a hybrid approach like EMT where prompts are used to elicit and reinforce specific responses, and a response is required from the child. In social learning theory, the clinician models and motivates language use (Fey, 1986; Scherer & Kaiser, 2010). Social learning theory has influenced the less intrusive approaches but also a hybrid approach like FS.

None of the above-mentioned language learning theories account for the difference between the children who acquire language effortlessly and those who struggle in doing so. Hence, carefully designed and thoroughly executed intervention studies are needed to determine how best to facilitate learning in children who do not develop adequate communication skills. Although effect of early intervention is well-documented (Law, Garrett, & Nye; Roberts & Kaiser, 2011; Tosh et al., 2017), in particular for measures of expressive language, only a handful of experimental studies have investigated the effects of early intervention in toddlers with CP (Broen, Doyle, & Bacon, 1993; Scherer, 1999; Scherer et al., 2008b; Ha, 2015; Kaiser et al., 2017).

DESIGNING INTERVENTION STUDIES

INTERVENTION DESIGN

Conducting early intervention studies is a complex affair, and there are numerous sources of bias and confounding factors that can affect outcomes. The design of an intervention study is fundamental to the validity of results. The randomised controlled trial (RTC) involving large number of participants is considered to be the most rigorous method of determining whether there is a cause-effect relationship between intervention and outcome because the RTC, when well-conducted, displays the lowest risk of bias and confounding (Irwin, Pannbacker, & Lass, 2008). However, large scale RTCs are not always feasible or even desirable when conducting intervention studies within the field of speech-language pathology. Randomisation may be difficult to achieve, especially when non-treatment control groups are involved and random group assignment may be considered unethical. Involving large numbers of participants increases reliability and generalisability, but small clinical groups may prevent inclusion of large participant numbers. Furthermore, information of individual developmental traits may be lost in larger studies. This information may be particularly important in small heterogeneous clinical populations. And finally, large studies are very costly. Thus, it may be more sensible to conduct smaller scale studies while maximising the level of experimental control (Ebbels, 2017).

Parent-implemented intervention introduces yet another confounding factor. Parent-implemented intervention is a triadic model where the SLP teaches the parent to use language support strategies in parent-child interaction. The effect of the intervention is dependent on parents' ability to learn these strategies. Thus, it is necessary to apply a fidelity measure to control for the effect of the parents' ability to use these strategies to a certain criterion level (Roberts & Kaiser, 2011).

SELECTION OF PARTICIPANTS

Experimental control in intervention studies can be increased by using a control group that does not receive intervention. However, selection bias may occur when participants are not randomised (Shaw et al., 2016). Participants may be selected based on the particular school they attend, their parents may have enrolled them in a speech-language therapy program in a certain university clinic, or their parents may decline participation for reasons that may systematically bias results (e.g. low socio-economic status). Thus, when randomisation is not an option, great care should be taken in matching the control group as closely as possible to the experimental group on measures that could potentially influence outcomes. Further, eligibility procedures should always be described so that it can be determined whether participants are actually representative of the clinical group they belong to. In toddlers, general development (including both maturation and learning) is expected and may explain changes in speech-language performance over time. Hence, pre-intervention measures such as age and speech-language performance are important for matching groups. If groups are very similar at pre-test, it is likely that greater progress in the experimental group than in the control group is caused by the intervention as matching controls for the effect of general development (Ebbels, 2017).

The theoretical rationale behind speech-language intervention studies in toddlers is that it is possible to positively affect speech-language development through intervention, and that this will have positive implications for later speech-language development. However, unless all toddlers are offered intervention, irrespectively of speech-language status, it is necessary to identify toddlers that are at risk of later speech-language difficulties. In general, it is harder to predict later difficulties the younger the child is, and predictors reported at group level cannot be assumed to apply to the individual child (Dollaghan & Campbell, 2009; Rescorla, 2011). Further, standardised tests of speech-language performance in toddlers below age 2 are scarce and in small languages, like Danish, non-existent. Nonetheless, it is important to define an accurate measure for distinguishing between toddlers whose speech-language status indicates need for intervention and toddlers who do not need intervention and to identify a clinical reference standard to which this measure can be compared (Dollaghan, 2004).

OUTCOME MEASURES AND ASSESSMENT

Outcome measures must be reflective of the research questions and sensitive to the intervention as well as clinically relevant and important to families. It may further be relevant to assess generalisation of outcome to other areas of speech or language or to other situations (Ebbels, 2017). Long-term results are also desirable to determine whether intervention effects are maintained, diminished, or perhaps even improved after termination of intervention. As a minimum, pre- and post-test assessment should be performed before and after intervention to evaluate effects of the intervention. Assessments should be carried out by the same tester in the same situation, and outcomes should be evaluated in the same way at both pre- and post-test. To reduce the risk of bias, the tester should be blinded to participant group allocation, both during assessment and evaluation (Irwin et al., 2008).

Speech-language assessment of toddlers poses special challenges. Toddlers may not say much during assessment, and it may thus be difficult to obtain a sample that is representative of the toddler's speech-language abilities. Parents may be used as assessors as parents have the advantage of spending more time with their child and hence can provide estimates that are not dependent on assessment performance. In particular, the CDI parent questionnaire can provide valid and reliable estimates of productive vocabulary. Assessment of observed productive vocabulary relies on identification of word use in spontaneous vocalisations, but identification may be limited when toddlers are in the early stage of word learning where prelinguistic and meaningful vocalisations are not easily distinguished, or their word productions may not bear much resemblance to adult targets (Vihman & McCune, 1994; Stoel-Gammon, 1989). Therefore, establishing criteria for word identification are important. Assessment of consonant inventory should include both prelinguistic and meaningful vocalisations because vocabularies are often small and may not represent the entire range of consonants a young child is able to produce (Stoel-Gammon, 1989).

Another challenge in speech-language assessment of toddlers is comparison across individuals. Spontaneous speech-language samples of the same length may contain very different amounts of vocalisations. For instance, Van Severen, Van Den Berg, Molemans, and Gillis (2012) found that consonant inventory increased with sample size. Hence, even though a specific consonant was present in one very voluble child, it could not be determined whether a less voluble child was not able to produce this consonant, or simply did not produce enough utterances for this consonant to be present in the speech sample. On the other hand, basing the size of a speech-language sample on the speech-language unit to be measured may not capture the variability of the more voluble toddlers, e.g. when a toddler takes interest in a certain toy and repeats the same word or sound over and over again, variability will be reduced. Thus, speech-language assessment in toddlers involves a compromise between valid group comparisons and retaining information from individual participants.

EARLIER STUDIES OF INTERVENTION IN TODDLERS WITH CLEFT PALATE

A systematic review of early language intervention in children with CP aged 1;6 to 4;11 years (Meinusch & Romonath, 2011) found limited empirical support for any intervention and recommended larger and better defined study groups, more transparent study designs, and inclusion of control groups of children, with or without CP, who also demonstrate need for intervention.

Five experimental studies have investigated the effect of early intervention on speech and language outcomes in toddlers (aged 12 to 36 months) with CP (Broen et al., 1993; Scherer, 1999; Scherer, D'Antonio, & McGahey, 2008b; Ha, 2015; Kaiser, Scherer, Frey, & Roberts, 2017) (see table 2). In total, thirty-nine toddlers with CP received intervention.

Broen et al. (1993) described a case of a 33-months-old girl with CP and age-typical language skills, limited production of oral consonants, and suspected velopharyngeal dysfunction (VPD). Her mother was trained to use word targets with incorporated consonant targets with the aim of facilitating consonant place features. Outcomes were increased accuracy of labial and alveolar place and increased glottal productions. This study provided a detailed description of the intervention and valuable clinical insights but did not attempt to control for the effect of general development.

Table 2. Characteristics of earlier studies of early intervention in toddlers with CP.

	Broen et al., 1993	Scherer, 1999	Scherer et al., 2008	Ha, 2015	Kaiser et al. 2017
N int group	1	3	10	17	8
N cont group	None	None	10 children w/o CP	9 children with CP	11 children with CP
Age in months (mean int, mean cont)	33	24-28 (25.3)	15-35 (27.4, 20.2)	15-29 (19.5, 16.4)	15-36 (24.3, 26.6)
Study design	Single-case	Single-case series	Quasi-exp group	Quasi-exp group	Small RCT, stratified for gender and age
Intervention	Parent-implemented FS	Clinician-implemented EMT	Parent-implemented FS	Parent-implemented naturalistic intervention	Clinician-implemented EMT
Setting	Home	Clinic	Clinic/home	Clinic/home	Clinic
Dose frequency and session duration:					
- Intervention	Weekly sessions with SLP	12-15 biweekly sessions of 50 min	2-4 sessions of 45 min with SLP	1 session with SLP for 3-4 hours, bi-weekly coaching via phone	48 biweekly sessions of 30 minutes
- Homework	1-2 times/day for 10 min	None	Unknown	Unknown	None
Total intervention duration	11 months	Unknown	Post-test after 3 months	3 months	Appr. 6 months

EMT=enhanced milieu teaching, FS=focused stimulation

Scherer (1999) included three boys with expressive language delays and restricted phonetic inventories aged 24-28 months in a study with a multiple baseline design. Target words included words that the participants comprehended but did not yet produce as well as words they did not comprehend. These target words incorporated word-initial consonants both within and outside the participants' pre-test inventories. Results showed that participants were able to learn most words, but that comprehended words were acquired faster than not comprehended words. There was also a slight tendency for words beginning with consonants within-inventory to be acquired faster than words with outside-inventory consonants. Participants increased their reported and observed productive vocabularies as well as their consonant inventories from pre-test to post-test. Nonetheless, whereas use of target words was monitored at multiple time points, the outcome measures, including vocabulary and consonant inventory measures, were not. Thus, there was no control of the effect of general development. It is also worth noticing that stable baselines were not obtained for all target words, which means that participants began to use these target words before they were introduced in intervention. This demonstrates the fast pace of general development in toddlers and points to the challenge of employing single-subject multiple baselines or reversal designs in this age group.

Scherer et al. (2008b) investigated the effect of training parents to use word targets with incorporated consonant targets on ten toddlers with CP aged 18-35 months compared to a control group of ten toddlers without CP aged 15-30 months. Recruitment procedures were described, and fidelity measures for parents' use of support strategies were reported. However, eligibility procedures were not reported. General inclusion criteria were applied, but speech-language inclusion criteria were not. Thus, it is not possible to determine if the participants with CP were representative of the general population of toddlers with CP and speech and/or language delay. Similar gains in productive vocabulary, consonant inventory, and speech accuracy were reported for both groups. Hence, these measures did not increase beyond the effect of general development. However, group comparison should be cautioned in this study. Although groups were matched on reported productive vocabulary, participants in the control group had age-typical speech-language development and were on average seven months younger than the toddlers with CP who for the most part had small vocabularies for age. Thus, although the groups were matched on productive vocabulary size, they were not matched on productive vocabulary size for age. In addition, productive vocabulary development is likely to follow a different trajectory in toddlers with CP compared to typically developing toddlers and therefore, toddlers with CP should ideally be compared to other toddlers with CP in intervention studies targeting productive vocabulary.

Ha (2015) investigated the effect of parent-implemented naturalistic intervention with no specific speech or language targets in 17 Korean-learning toddlers with CP aged 15-29 months compared to a control group of nine toddlers with CP aged 13-23 months. Fidelity measures for parents' use of support strategies were reported, but recruitment and eligibility procedures were not. General inclusion criteria were applied, but speech-language inclusion criteria were not. Hence, it is also unclear from this study whether the participants were representative of the general population of toddlers with CP and speech and/or language delay. Korean CDI norms suggest that toddlers in both groups had age-typical reported vocabulary scores (Frank, Braginsky, Yurovsky, & Marchman, 2017). Nonetheless, the toddlers in the control group were on average three months younger than the intervention group and had a lower reported productive vocabulary size (reported vocabulary mean: 8.2 vs. 16.1). As groups were not equivalent at pre-test, the reported effects on productive vocabulary measures and MLU should be interpreted with caution.

Most recently, Kaiser et al. (2017) reported results from a small RCT of 19 toddlers with CP aged 15-36 months. Eight toddlers were included in the intervention group while 11 toddlers received business-as-usual treatment. Target words incorporated word-initial consonants both within and outside the participants' pre-test inventories. Participant recruitment procedures were described, but eligibility procedures were not, and it was not explained why the randomised design yielded different numbers of participants in the two groups. General inclusion criteria were applied as well as speech-language inclusion criteria. However, the language inclusion criterion was a reported vocabulary of at least five words, whereas a diagnosis of speech or language impairment was not required. American CDI norms indicate that toddlers in both groups on average had CDI percentile scores well within the normal range (Fenson et al., 2007) and thus were probably not language-delayed. The speech inclusion criteria were: compensatory error on at least one phoneme, or consonant inventory of fewer than five stop or nasal consonants in all positions, or errors on at least two stop or nasal consonants. Nevertheless, the predictive value of these criteria in children with CP is unclear. Moreover, meeting these criteria would be highly influenced by age-appropriate developmental errors and by sample size as the speech measures were obtained from a naming test and it was not reported how many words the participants named. Thus again, the representativeness of participants cannot be determined. Significant group differences were found in reported productive vocabulary gain, PPC-A gain, and gain in receptive language scores from pre-test to post-test. However, participants in the intervention group had considerably lower scores at pre-test than the control group (reported vocabulary mean: 182.0 vs. 303.9; PCC-A mean: 31.8 vs. 45.7; receptive language mean: 87.4 vs. 104.9). Hence, in spite of random group allocation, groups were not equivalent at pre-test and the reported effects should thus be interpreted with caution.

In summary, earlier intervention studies involving toddlers with CP have not completely accounted for participant selection procedures and have rarely employed speech-language inclusion criteria. This means that it is difficult to determine if participants in these studies were in fact speech-language delayed and thus representative of toddlers with CP and speech-language delay in general. In addition, these earlier studies have not included appropriate control for the effect of general development, and experimental groups and control groups have not been sufficiently matched. Hence, interpretation of these studies' child outcomes should be cautioned.

OVERVIEW OF STUDIES AND PARTICIPANTS

This section presents the main research questions and hypotheses as well as an overview of the three studies of the dissertation. Further, the participants and their distribution across studies are described.

The central focus of this dissertation is whether early delays in vocabulary and/or speech development in toddlers with CP can be positively affected by early intervention. As it is well-established that some children with CP will never need intervention, it is further necessary to define what constitutes delayed vocabulary and speech development in toddlers with CP and to develop a time-efficient procedure for evaluation of these measures. Hence, study 1 aims to develop and validate a clinically useful screening procedure of consonant inventory and vocabulary for toddlers with CP in order to distinguish toddlers in need of early intervention from toddlers who do not need intervention. The first research question is whether trained raters can reliably assess consonant inventory in toddlers with CP using real-time listening (RTL) assessment. The hypothesis is that RTL will be a reliable assessment procedure in toddlers with CP because it has been reported that trained raters can reliably assess prelinguistic utterances in younger children both with and without CP using RTL assessment, and because toddlers with CP as a group are expected to exhibit delayed vocabulary development and restricted consonant inventories and thus still have many utterances that are either prelinguistic or unintelligible. The central research question is how well the combined information from RTL and the CDI in the screening procedure identifies need for intervention compared to a clinical reference standard of experienced SLPs' clinical evaluations. The hypothesis is that there will be substantial agreement between the two evaluation procedures because the screening procedure is based on possible early predictive measures of later speech-language difficulties in toddlers with CP, and it has been reported that at least one of these predictive measures (oral stop production) may form the basis for expert SLPs' evaluation of intervention need.

To investigate whether early delays in vocabulary and/or speech development in toddlers with CP can be positively affected by early intervention, studies 2 and 3 explore the effects of parent-implemented FS intervention in toddlers with CP and delayed speech and/or language development compared to a matched control group of toddlers with CP. Study 2 focuses on short-term results while study 3 is concerned with long-term outcomes.

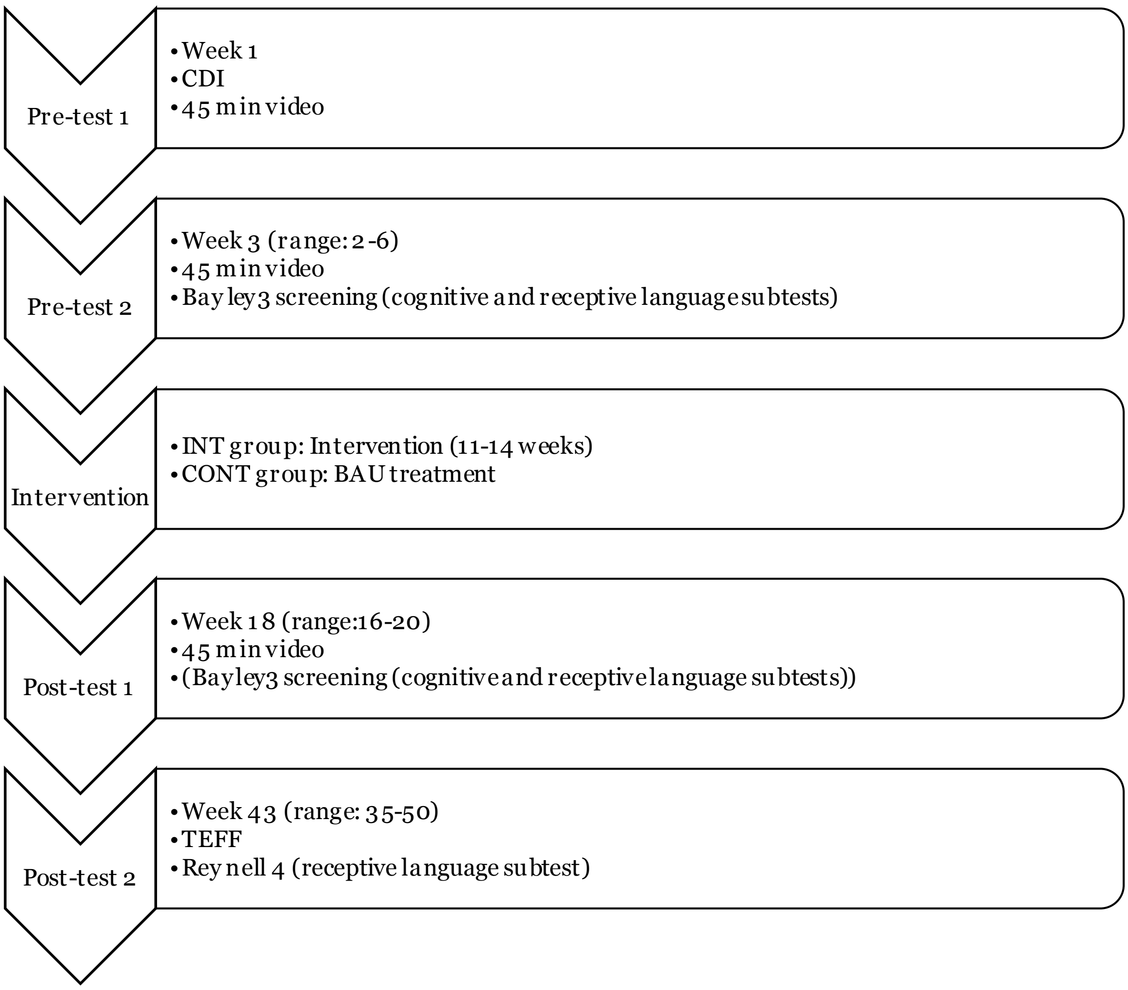
The central research question in study 2 is whether measures of observed productive vocabulary and consonant inventory in toddlers with CP and delayed speech and/or language development are positively affected by parent-implemented FS intervention in which vocabulary and consonant inventory are directly targeted. One hypothesis is that vocabulary will expand to a greater degree in the intervention group than in the control group. This effect has been reported in numerous studies of parent-implemented early intervention in other clinical populations and is based on the observation that parent-child interaction involving use of language support strategies can positively affect child language outcomes. A second hypothesis is that consonant inventory will expand as a function of vocabulary expansion. This hypothesis is driven by the observation that vocabulary and consonant inventory development are closely interlinked in the early vocabulary. It is assumed that this link will be aided by including word targets with consonants both within and outside the toddler's pre-test consonant inventory, and by teaching parents speech recasting to facilitate comparison between child production and adult phonological target form.

The central research question in study 3 is whether parent-implemented FS intervention, targeting vocabulary and consonant inventory directly, has long-term effects on measures of speech accuracy, reported productive vocabulary, and receptive language in toddlers with CP and delayed speech and/or language development. The main hypothesis is that the intervention group will perform better than the control group on all productive outcome measures as the underlying tenet of home-based parent-implemented intervention is that effects are expected to maintain over time because parents are taught to use language support strategies in naturally occurring interactions with their child. This hypothesis is based on the assumption that toddlers in the intervention group will expand their consonant inventories to a larger degree than toddlers in the control group, and the observation that toddlers both with and without CP with larger consonant inventories attain higher speech accuracy scores. In addition, because FS is based on a transactional perspective of language acquisition, it may be assumed that parents' responsiveness and contingent language input help toddlers in the intervention group to infer the meaningful relations between the natural context and words. Thus, another hypothesis is that the intervention group will have higher receptive language scores than the control group. Finally, it is hypothesised that more toddlers in the intervention group compared to the control group will perform as well as peers without CP on measures of speech accuracy because toddlers in the intervention group are expected to perform better on measures of speech accuracy than toddlers in the control group. Table 3 provides an overview of the focus, design, and participants in the three studies, and figure 2 provides an overview of the design of the total project and procedures.

Table 3. Focus, design, and participants in the included studies.

Study	Focus	Design	Participants
1	Development and validation of a speech-language screening tool for toddlers with CP	Cross-sectional	22 toddlers with CP
2	Evaluation of short-term intervention effects on measures of productive vocabulary, consonant inventory, and need for intervention	Longitudinal	28 toddlers with CP: 14 in the intervention group and 14 in the control group
3	Evaluation of long-term intervention effects on measures of speech accuracy, productive vocabulary, receptive language, and need for intervention	Longitudinal	26 toddlers with CP: 13 in the intervention group and 13 in the control group

Figure 2. Project design and procedures.



PARTICIPANTS AND THEIR DISTRIBUTION ACROSS GROUPS AND STUDIES

A total of 55 toddlers with CP were included in this project. All participants had operated CP, at least one native Danish-speaking parent, and absence of a known genetic syndrome, severe cognitive delay, or sensorineural hearing loss. Table 4 shows characteristics of all 55 participants.

Participants' age ranged from 16-24 months at pre-test (mean age: 19.11 months). Thirty participants were boys and 25 were girls. Thirty-seven had cleft lip+cleft palate (CLP) whereas 18 had cleft palate only (CPO). The distribution of genders within cleft type is illustrated in figure 3. CLP was more prevalent compared to CPO in this study than in the total clinical population, but the distribution of genders within cleft type reflected the distribution in the total clinical population.

Figure 3. Distribution of gender within cleft type

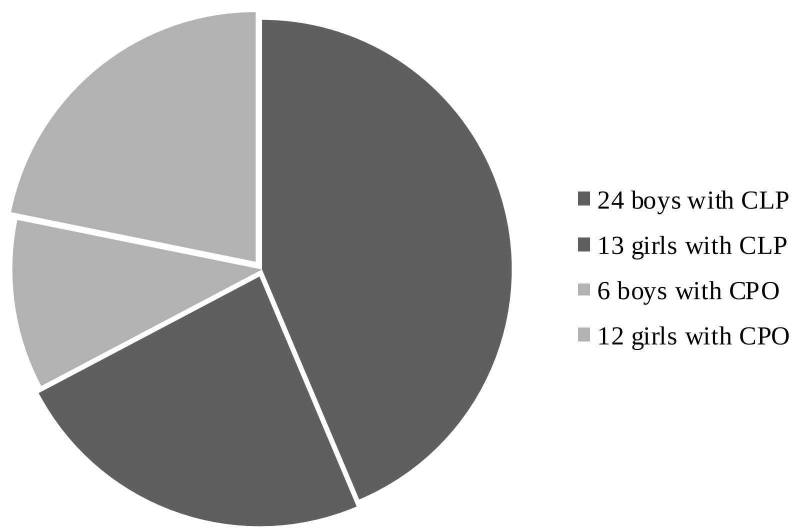


Table 4. Participant characteristics.

Child	Center	Study 1	Study 2	Study 3	Gender	Cleft type	SES	Age in months at pre-test	Age at palatal surgery	CDI vocabulary at pre-test	CDI percentile at pre-test	Consonant inventory at pre-test (screening)
1	C	19	1 INT	1 INT	M	UCLP	4	24	14	17	0	j m n h*
2	C	20	2 INT	2 INT	M	UCLP	2	24	14	25	5	w j m n h*
3	C	12	3 INT	3 INT	M	UCLP	4	20	14	40	25	b̥ g̥ w j m n h*
4	C	10	4 INT	4 INT	M	CPO	3	20	14	22	10	b̥ d̥ g̥ tˢ kʰ f s w j m n ŋ h*
5	C	15	5 INT	5 INT	M	BCLP	3	18	12	9	5	b̥ l j m h*
6	C	11	6 INT	6 INT	F	BCLP	2	17	12	5	2.5	d̥ g̥ w j m n h*
7	C		7 INT	7 INT	M	UCLP	4	20	14	34	25	b̥ g̥ w j m n ŋ h
8	C		8 INT	8 INT	M	UCLP	4	20	13	6	0	b̥ g̥ w j m n h
9	C		9 INT	9 INT	M	UCLP	2	18	14	4	5	d̥ g̥ s j m n ŋ h
10	C		10 INT		M	CPO	2	18	6	0	0	b̥ d̥ g̥ x w j m ŋ h
11	C		11 INT	11 INT	M	UCLP	4	20	16	20	10	w j m n ŋ h
12	C		12 INT	12 INT	M	UCLP	2	18	14	5	2.5	b̥ d̥ w j m n ŋ h
13	C		13 INT	13 INT	M	UCLP	2	17	12	5	10	b̥ w j m ŋ h
14	C		14 INT	14 INT	M	CPO	3	17	13	5	10	w j m n ŋ
15	A	22	1 CONT	1 CONT	M	BCLP	3	20	13	10	2.5	w j m n h ʔ*
16	A		2 CONT	2 CONT	F	CL/SMCP	3	21	-	11	2.5	tˢ w l j m n h
17	A	18	3 CONT	3 CONT	M	UCLP	3	17	12	8	20	w j m ŋ h*
18	A		4 CONT	4 CONT	M	CPO	2	19	16	13	5	b̥ d̥ g̥ tˢ kʰ s x w l j m n ŋ h
19	A		5 CONT	5 CONT	F	UCLP	4	19	14	18	5	b̥ g̥ w j m n h
20	A		6 CONT	6 CONT	F	CPO	5	18	16	9	2.5	b̥ d̥ g̥ w j m n ŋ h
21	A		7 CONT	7 CONT	F	CPO	4	18	15	21	30	w j m n h
22	A		8 CONT	8 CONT	M	UCLP	4	18	14	0	0	b̥ d̥ g̥ x w l j m n h
23	A	21	9 CONT	9 CONT	F	CPO	5	19	14	11	2.5	w j m n h*
24	A		10 CONT	10 CONT	M	UCLP	4	17	14	5**	0**	j m h
25	A		11 CONT	11 CONT	M	UCLP	2	19	14	14	5	w j m n h
26	A	16	12 CONT		M	CL/SMCP	4	18	-	5	2.5	b̥ m h*
27	A		13 CONT	13 CONT	M	UCLP	4	16	14	4	5	w j m n h

28	A		14 CONT	14 CONT	M	UCLP	4	16	12	16**	5**	w j m h
29	C	1			F	CPO	2	23	14	156	40	b̥ d̥ g̥ p^h t^s k^h s w j m n ŋ h*
30	C	2			F	CPO	3	17	12	21	30	b̥ d̥ g̥ w j m n ŋ h*
31	C	3			F	CPO	3	23	12	288	65	b̥ d̥ g̥ p^h t^s k^h f s w j m n h*
32	A	4			F	UCLP	4	24	16	110	15	b̥ d̥ g̥ p^h k^h s w l j m n ŋ h*
33	A	5			F	CPO	3	17	12	23	35	b̥ p^h t^s w l j m n h*
34	A	6			F	CPO	2	25	9	297	50	b̥ d̥ g̥ t^s k^h f s w l j m n ŋ h*
35	A	7			F	UCLP	3	21	13	85	25	b̥ d̥ g̥ t^s k^h f s w j m n ŋ h*
36	A	8			F	CPO	3	22	13	87	75	b̥ d̥ t^s f s w l j m n *h
37	A	9			F	CL/SMCP	3	23	-	70	15	b̥ d̥ g̥ k^h f s w j m n ŋ h*
38	C	13			F	BCLP	2	23	13	143	35	g̥ w j m n ŋ h ʔ*
39	C	14			M	BCLP	2	22	14	169	55	b̥ g̥ s w j m n ŋ h*
40	A	17			M	UCLP	3	23	12	133	40	w j m n ŋ h ʔ*
41	A				F	BCLP	5	22	14	59	10	b̥ g̥ f s w j ʁ m n ŋ h
42	A				M	UCLP	-	19	14	46	60	b̥ d̥ g̥ k^h w j m n ŋ h
43	A				F	CPO	5	19	12	139	80	b̥ d̥ g̥ p^h f s w l j m n h
44	A				F	UCLP	2	17	13	39	60	b̥ d̥ g̥ w j m n ŋ h
45	A				F	BCLP	3	19	14	130	55	b̥ d̥ g̥ k^h x w l j m n h
46	C				F	CPO	1	18	6	17	20	b̥ d̥ g̥ f s w j m n h
47	A				M	CPO	2	17	6	61	90	b̥ d̥ g̥ w j m n h
48	A				F	CPO	2	16	14	18	50	b̥ d̥ g̥ s w l j m n ŋ
49	A				M	CPO	3	15	14	15	45	b̥ d̥ s w j m n ŋ h
50	C				F	CPO	4	17	12	14	20	b̥ d̥ t^s f w j m n ŋ h
51	C				M	CL/SMCP	4	16	-	21	55	b̥ d̥ g̥ p^h k^h w l j m n h
52	C				M	BCLP	2	16	12	8	20	d̥ w l j m n ŋ h
53	C				F	UCLP	3	17	13	16	15	b̥ d̥ g̥ p^h t^s k^h s w l j m n h
54	C				F	BCLP	5	17	14	18	25	b̥ d̥ g̥ w j m n h
55	C				M	BCLP	-	17	13	57	80	g̥ w j m n ŋ h
56***	C				F	UCLP	5	22	19	6	0	g̥ w j m n ŋ h

*Inventory agreed by two out of three raters in Study 1. **Scores are from post-test after four months. Scores from pre-test were not available.

***Participant excluded after screening because of severe illness. C=Copenhagen cleft palate centre, A=Aarhus cleft palate centre.

It is no secret that well-educated middle-class parents are more likely to enrol their children in scientific studies. This tendency is also to some degree reflected in this project. Table 5 shows the percentage distribution of social classes in the general Danish population (Ploug, Andersen, Olsen, & Juul, 2012) and in the participating families of this project. In this project, there are more higher middle class families and fewer working class families than in the general population whereas the distribution of higher class, lower middle class, and lower class families is fairly similar to the general population. In two families, both parents were students and thus socio-economic status[5] (SES) could not be calculated.

Table 5. Distribution of social classes in the general population and in the present project.

	Higher class	Higher middle class	Lower middle class	Working class	Lower class
General population	2%	12%	30%	42%	14%
Families in this project	1.9% (n=1)	30.2% (n=16)	30.2% (n=16)	28.3% (n=15)	9.4% (n=5)

Participants were recruited from a complete birth cohort of all 103 Danish children with CP born between December 2012 and August 2014. Figure 4 illustrates the eligibility procedures of the entire project. The participants were distributed into different groups based on their speech-language performance and whether they were seen at the cleft palate centre in Copenhagen or Aarhus. Speech-language criteria for need for intervention were a productive vocabulary score at or below the 10^{th} percentile on the CDI, and absence of alveolar stops. In addition, participants must produce below 50 words on the CDI to be included in the intervention group. Information on group allocation and distribution across studies are provided in table 6.

Group allocation was based on where the families lived as the author, who carried out all intervention, was situated in Copenhagen. However, random group allocation would have raised ethical concerns. The present design allowed parent information to specifically state that families in the Aarhus centre would act as controls whereas families in the Copenhagen centre would receive intervention if they needed it. Further, families in the control group were allowed to seek local speech therapy services. Three toddlers from the Copenhagen centre needed intervention based on a productive vocabulary score at or below the 10^{th} percentile on the CDI, and/or absence of alveolar stops, but their productive vocabularies exceeded 50 words and thus, these toddlers were not eligible for the intervention group. However, these toddlers

[5] SES is based on highest level of education and current employment status (Ploug, Andersen, Olsen, & Juul, 2012).

still received intervention as it would have been unethical to ignore their intervention need. Intervention outcomes for these toddlers are not reported in this dissertation.

Figure 4. Flowchart of the eligibility procedures of the entire project.

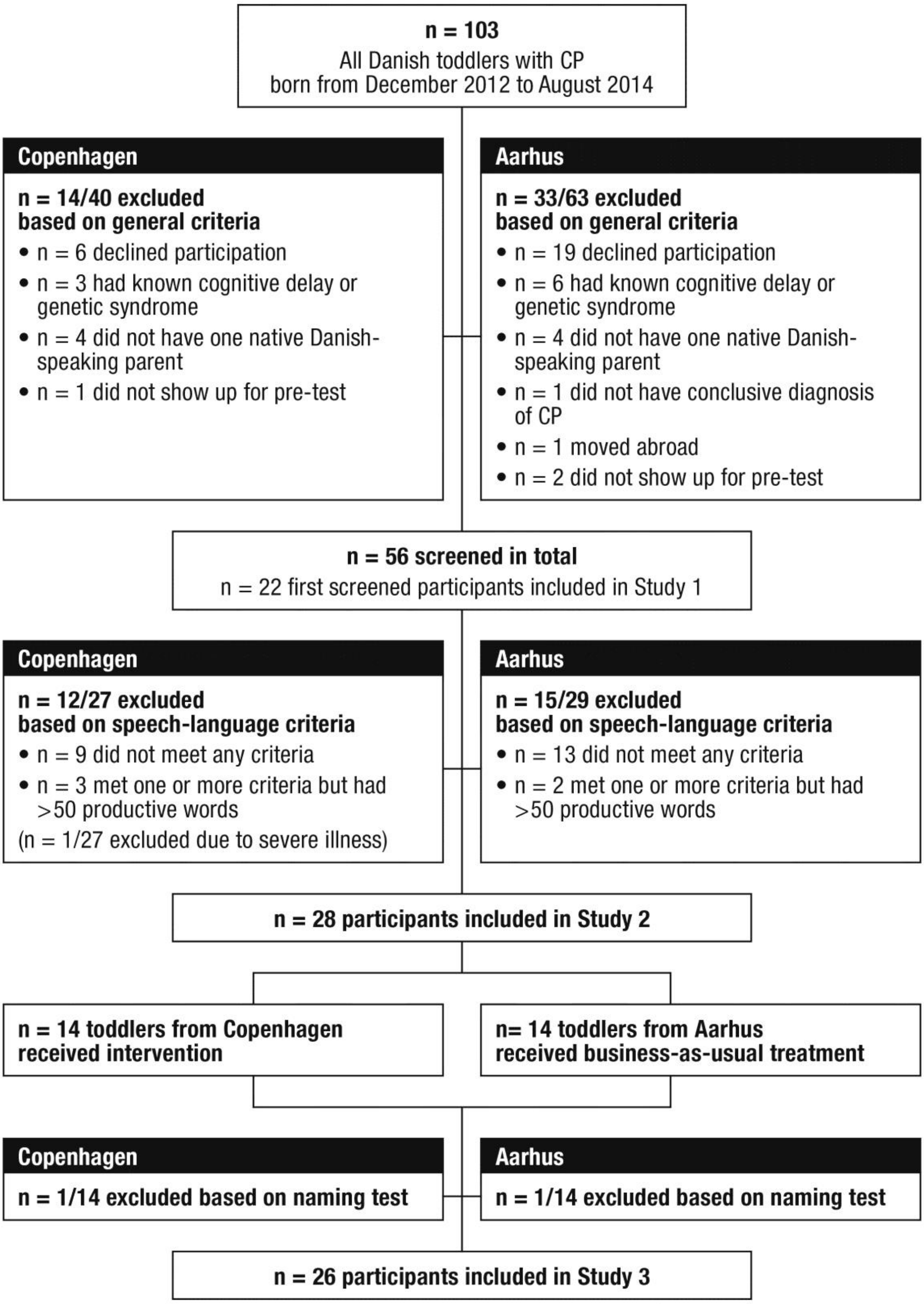


Table 6. Group allocation and distribution of participants across studies.

Total # participants	Speech-language inclusion criteria	Group # participants	Study # participants
Copenhagen # 26	</=10th percentile CDI productive vocabulary AND/OR no alveolar stops <50 CDI productive words	+Need for intervention (INT) group # 14	Study 1 # 6 Study 2 and 3 # 14 and # 13
	</=10th percentile CDI productive vocabulary AND/OR no alveolar stops >50 CDI productive words	+ Need for intervention, large vocabulary # 3	Study 1 # 2
	>10th percentile CDI productive vocabulary AND alveolar stops >50 CDI productive words	- Need for intervention # 9	Study 1 # 3
Aarhus # 29	</=10th percentile CDI productive vocabulary AND/OR no alveolar stops <50 CDI productive words	+ Need for intervention (CONT) group # 14	Study 1 # 4 Study 2 and 3 # 14 and # 13
	</=10th percentile CDI productive vocabulary AND/OR no alveolar stops >50 CDI productive words	+ Need for intervention, large vocabulary # 2	Study 1 # 1
	>10th percentile CDI productive vocabulary AND alveolar stops >50 CDI productive words	- Need for intervention # 13	Study 1 # 6

STUDY 1

Development and validation of a screening procedure to identify speech-language delay in toddlers with cleft palate

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Abstract
The purpose of this study was to develop and validate a clinically useful speech-language screening procedure for young children with cleft palate ± cleft lip (CP) to identify those in need of speech-language intervention. Twenty-two children with CP were assigned to a +/– need for intervention conditions based on assessment of consonant inventory using a real-time listening procedure in combination with parent-reported expressive vocabulary. These measures allowed evaluation of early speech-language skills found to correlate significantly with later speech-language performance in longitudinal studies of children with CP. The external validity of this screening procedure was evaluated by comparing the +/– need for intervention assignment determined by the screening procedure to experienced speech-language pathologist (SLP)s' clinical judgement of whether or not a child needed early intervention. The results of real-time listening assessment showed good-excellent inter-rater agreement on different consonant inventory measures. Furthermore, there was almost perfect agreement between the children selected for intervention with the screening procedure and the clinical judgement of experienced SLPs indicate that the screening procedure is a valid way of identifying children with CP who need early intervention.

Keywords: Cleft palate, cleft palate speech, early intervention, speech-language screening

Introduction
Children with CP may exhibit delay in speech sound development, phonological/articulatory delay/disorder, hypernasality, expressive language delay/disorder, and decreased intelligibility (Chapman & Willadsen, 2011; Kuehn & Moller; 2000). Despite decades of debate and research on optimal surgical methods and timing (Leow & Lo, 2008; Peterson-Falzone, Hardin-Jones, & Karnell, 2010), the prevalence of speech and language delay/disorder is still relatively high in this population.

Prevalence and consequences of speech-language delay in children with CP
In a large cross-sectional study of 1100 five-year-olds with CP (Britton et al., 2014), 50% of the children had speech within the normal range, defined by cut-off scores for nasality and cleft speech characteristics. Similarly, a literature review of speech outcomes in children with CP (Lohmander, 2011) found that depending on type and extent of the cleft, 50-60% of 3-year-olds and 60-70% of five-year-olds had 'good speech', although this criterion was not further defined due to the heterogeneous speech outcome measures in the included articles. In young children, language is important for learning, engaging in social relationships, and regulation of behaviour and emotion (Cohen, 2010). Albeit the consequences of speech-language delay in toddlers with CP on later socio-emotional outcomes have not been investigated, one longitudinal study (Havstam, Sandberg, & Lohmander, 2011) has linked disordered speech in

5-year-olds to negative attitude towards communication in 10-year-olds, even when the speech disorder was resolved. Furthermore, toddlers with speech-language delay/disorder are at risk of developing later expressive language delay (Rescorla, 2011), and speech proficiency correlates with both conversational style and early reading skills in children with CP (Chapman, 2011; Frederickson, Chapman, & Hardin-Jones, 2006). Offering appropriate speech-language intervention as soon as possible, therefore, seems to be pertinent, and hence, early identification of children with speech-language delay is essential. Conversely, it is important to avoid unnecessary intervention which adds to the burden of care for the family and taps into society's limited resources for speech-language intervention.

Need for early intervention in children with CP

Children's need for speech-language intervention is typically determined by an SLP and based on information from parents and the SLP's observation of the child within a limited time frame. The results from a retrospective study (Hardin-Jones & Chapman, 2008) show that children with CP who were referred to early speech intervention by an SLP in a CP clinic at 17 months had poorer speech-language outcomes at 27 months than children with CP who were not referred for intervention. Although this unfortunately means that the speech-language intervention provided to the children in this study was not always sufficiently successful, it also suggests that specialist SLPs in cleft palate clinics are able to distinguish between children with CP who need intervention, and those who do not. However, not all children with CP have access to specialist SLPs, and SLPs need training before they become specialised. Even when children with CP do have access to specialised care, not all centres focus on early intervention, meaning that SLPs might lack knowledge of early speech-language development and red flags for delayed development in these children. Therefore, a screening procedure for determining which children need intervention will be clinically useful for SLPs without expertise in children with CP and for training future experts.

A screening procedure may also be important to researchers. In one single-case study of early intervention in a toddler with CP (Broen, Doyle, & Bacon, 1993), the participant was described as having normal language development, but enrolled in intervention due to lack of oral consonant production. In a second study of early intervention in three toddlers with CP, Scherer (1999) reported participants' language scores from the Preschool Language Scales (Edition 3) as well as reported and observed expressive vocabulary, syllable types, mean length of utterance, and consonant inventories and concluded that all participants showed expressive language delay and restricted phonetic inventories. However, in the only three group studies published on early intervention in toddlers with CP (Blakeley & Brockman, 1995; Ha, 2015; Scherer, D'Antonio, & McGahey, 2008), speech and language performance was not part of the inclusion criteria that were: CP, normal cognition, and hearing above a certain threshold.

Inclusion of speech-language criteria would ensure that participants actually show an early delay in speech-language development and are at risk of continued speech and language difficulties. In addition, it will enable replication of studies, and it will provide a foundation for appropriate matching of control groups which is vital in this age group due to a strong maturation effect. At present, speech norms for children younger than 3 years do not exist (although a test of phonology for children around 2 years (Stoel-Gammon & Williams, 2013) is being developed). Furthermore, available language tests are difficult to undertake in toddlers and may not detect less severe language delays (Scherer, Frey, & Kaiser, 2014). Accordingly, a screening procedure based on 'red flags' for later speech-language difficulties may be a relevant addition to the participant selection process in future studies.

Correlation between early speech-language skills and later speech-language performance in children with CP

Longitudinal studies evaluating correlations between early speech-language skills and later speech and language performance offer the possibility of identifying children with CP at risk of later speech-language difficulties at an early age. In children without CP, there is a large overlap between sounds and syllable shapes in babbling and early speech sounds and vocabulary (see Stoel-Gammon, 1998a, b for a review). In children with CP, however, this relationship is less obvious. Before palatal surgery, speech sound development may appear delayed, yet, growth of consonant inventory will be evident following palatal surgery for many children with CP (Chapman, Hardin-Jones, & Halter, 2003; Grunwell & Russell, 1987, 1988; Grunwell & Russell, 1988; Jones, Chapman, & Hardin-Jones, 2003; O'Gara & Logemann, 1988; O'Gara, Logemann, & Rademaker, 1994; Russell & Grunwell, 1993). Curiously, two studies even find negative correlations between pre and postsurgical speech skills (Chapman, 2004; Scherer, Williams, & Proctor-Williams, 2008). Chapman (Chapman, 2004: 253) presumed that "it takes considerable time for either the positive effects of successful surgery and/or speech and language intervention or the negative effects of continued VPD to change the course of development". By contrast, studies investigating speech-language skills in children after palatal surgery have found significant positive correlations between earlier and later speech and language skills (Chapman, 2004; Chapman et al., 2003; Klintö et al., 2014; Lohmander & Persson, 2008; Willadsen, 2007). Thus, the following review of early speech-language skills correlating significantly with later speech-language performance (henceforth termed 'predictive measures') only includes measures obtained after palatal closure and measures obtained from children with complete palatal closure at the time of evaluation.

In a longitudinal study with18 participants, Klintö et al. (2014) reported three measures at 18 months that correlated significantly with percent consonants correct, adjusted (PCC-A) at 36 months, with large effect sizes. These were number of different oral consonants, total number of dental stops and total number of oral consonant tokens. Only consonants occurring at least twice in different utterances were counted. All participants had their soft palate closed at 4 months; half of them received hard palate closure at 12 months, and the other half still had an unrepaired hard palate at 36 months, but the correlations were only significant in the group that received hard palate closure at 12 months.

In addition, *number of different oral stops, total number of oral stops, total number of anterior stops,* and *total number of dental stops* at 18 months correlated significantly with PCC-A at 36 months but had low effect sizes. *Number of different true consonants* at 21 months correlated significantly with number of different words (NDW) at 39 months in a study by Chapman (2004). *Number of different emerging consonants* (consonants produced correctly in words at least twice, regardless of word position) at 21 months correlated significantly with number of different emerging consonants, number of different true consonants, and NDW at 39 months (Chapman, 2004). All 15 children in these studies (Chapman et al., 2003; Chapman, 2004) had complete palatal closure at 12-13 months.

Percentage of oral stops at 17 months was moderately but significantly correlated with number of different stable consonants (consonants produced correctly in at least 70% of word-initial target occurrences) and percentage of consonants produced correctly at 27 months in a study of 30 children with CP who had complete palatal closure between 7 and 17 months and 10 children without CP (Hardin-Jones & Chapman, 2008). The correlation was found for the total group of children, and the authors did not report if this correlation persisted when children without CP were excluded.

Observed expressive vocabulary (NDW) in ten minutes of child-caregiver interaction at 21 months correlated significantly with NDW at 39 months in the study by Chapman (2004). Word status was determined from contextual support, parental identification, and phonetic or segmental similarity to the adult target.

Considering the above-mentioned predictive measures, it seems possible to identify children with CP at risk of later speech and language difficulties before age 2. A summary of the predictive measures is shown in table 1. It should be pointed out that these predictive measures were obtained in US English and Swedish-learning children whereas the present study involves Danish-learning children. Notwithstanding, English, Swedish, and Danish are all Germanic languages with rather similar phonological systems; the main difference being that Danish and Swedish in general have simpler systems with fewer consonant phonemes (Basbøll, 2005; Riad, 2014). Similarities between languages have also been found in studies of early sound acquisition in children with CP compared to peers without CP. For example, children with CP acquiring US English, Swedish, or Danish produce fewer oral stops, more glottals, and fewer coronals than their peers without CP (Chapman, Hardin-Jones, Schulte, & Halter, 2001; Hutters, Bau, & Brøndsted, 2001; Willadsen & Albrechtsen, 2006; Lohmander, Olsson, & Flynn, 2011). Hence, it is likely that predictive measures obtained in US English and Swedish also apply to Danish.

Table 1. Predictive measures at 17-21 months correlating with speech-language performance at 27-39 months.

17-21 months	27 months	36-39 months	No. of children	Reference
no. of different true consonants		NDW	15	Chapman, 2004
no. of different oral consonants		PCC-A	9	Klintö et al., 2014
no. of oral consonants		PCC-A	9	Klintö et al., 2014
no. of different emerging consonants		emerging consonants, true consonant inventory, NDW	15	Chapman, 2004
percentage of oral stops[A]	number of different stable consonants, percentage of correct consonants		30 (40)	Hardin-Jones and Chapman, 2008
no. of dental stops		PCC-A	9	Klintö et al., 2014
observed vocabulary (NDW)		NDW	15	Chapman, 2004

[A]Correlation found in a mixed group of children with and without CP

Assessment of speech production in toddlers

Another important property of a screening procedure must be time-efficiency while still allowing a reliable and valid evaluation of a child's speech and language performance.

In the literature, early speech production has traditionally been evaluated by narrow phonetic transcription of child utterances during spontaneous interaction between child and caregiver. However, compared to typical adult speech, intra and inter-rater reliability is generally low in phonetic transcription of speech of young children and in speakers with speech sound disorders (Shriberg & Lof, 1991), both of which applies to young children with CP. Furthermore, phonetic transcription is a time-consuming and thus expensive procedure. Especially in (group) intervention studies, time is an important factor since analysis of speech data needs to be performed immediately after pre-test sessions so intervention can commence within the shortest possible time frame.

One such time-efficient method is real-time listening (RTL) which has been found to be a valid method of estimating infants' consonant repertoire compared to both caregiver report and phonetic transcription (Ramsdell, Oller, Buder, Ethington, & Chorna, 2012). RTL had high inter-rater reliability in a methodological study of toddlers with CP (Willadsen, 2016). In RTL, raters simulate parents' way of listening to their child's speech. Rather than assessing exact phonetic inventory, the purpose of RTL is to assess the functional phonological repertoire a child controls. The functional repertoire is recognised by caregivers and can be viewed as the foundation for caregivers' interpretation of child utterances as meaningful, eventually resulting in the child attaching meaning to a sound string: the negotiation of word meaning (Veneziano, 1988). Furthermore, phonetic transcription greatly overestimates the size of a child's consonant repertoire as compared to parental report (Ramsdell et al., 2012). Thus, RTL seems potentially useful for evaluating predictive measures of later speech-language performance in children with CP as part of a screening procedure. However, Ramsdell et al. (2012) mainly studied children in the prelinguistic stage whereas the children in the present study all produced at least a few words.

This study aimed to develop and validate a clinically useful screening procedure for young children with CP in order to determine which children need early intervention. Consonant inventories are established by RTL assessment, and predictive measures from the consonant inventories are used for assigning children to +/− need for intervention conditions. The external validity of the screening procedure is assessed by comparing the +/− need for intervention group assignment from the screening procedure to experienced SLPs' clinical judgement of whether or not a child with CP needs early intervention. The study addresses the following experimental questions:

(1) Can trained raters reliably assess consonant inventory, including predictive measures of later speech-language performance, in children with CP using RTL assessment?

(2) Do experienced SLPs' clinical judgements agree with the screening procedure as to which children need intervention?

Method

Participants

Twenty-two native Danish-speaking children with CP between 17 and 25 months participated. They were the first to enrol in a larger intervention study in which all Danish children born with CP in 2013 and the first half of 2014 (n = 104) were invited to participate. Exclusion criteria were presence of a syndrome or severe cognitive delay (eight children), and none of the parents being a native Danish speaker (eight children). Six children were excluded for other reasons (e.g. moving abroad), and 26 parents declined participation. A total of 56 children with CP participated in the larger study. Participants in the present study all had complete palatal surgery between 9 and 16 months (mean age 13 months), and all participants were seen for pre-testing at least two months after palatal surgery to allow for post-surgical catch-up of speech and language development (see table 2).

All children scored within the 'competent' or 'emerging' categories on the Bayley III screening subtest of cognitive skills. None of the children had a known genetic syndrome, severe cognitive delay, or sensorineural hearing loss. However, formal testing was not performed. For the subtest of receptive language skills, all but two children scored within the 'competent' or 'emerging' categories. The remaining two children refused to participate in the receptive language screening, and several children fell into the 'emerging' category because they refused to participate in some of the test items.

Characteristics of the participants are shown in table 2, including history of otitis media with effusion (OME) and history of pressure-equalising tubes (PETs) before 12 months and between 12–18 months. Information is based on parental report and review of medical records. The results from hearing tests were not available.

All parents signed a written consent form to take part in this study, complying with ethical principles of the Declaration of Helsinki.

Table 2. Participant characteristics.

Child	Gender	Age at palatal surgery	Age in months at evaluation	Cleft type	Bayley III-Cognitive screening	Bayley III-Receptive language screening	History of OME	PETs before 12 m/PETs 12-18 m
1	F	14	23	CPO	COMPETENT	COMPETENT	YES	YES/YES (one ear)
2	F	12	17	CPO	COMPETENT	COMPETENT	YES	NO/YES
3	F	12	23	CPO	COMPETENT	COMPETENT	YES	YES/NO
4	F	16	24	UCLP	COMPETENT	EMERGING	YES	NO/YES
5	F	12	17	CPO	COMPETENT	COMPETENT	UNKNOWN	NO/NO
6	F	9	25	CPO	EMERGING	EMERGING	YES	YES/NO
7	F	13	21	UCLP	COMPETENT	COMPETENT	YES	YES/NO
8	F	13	22	CPO	COMPETENT	EMERGING	YES	YES/YES
9	F	-	23	CL/SUBM	COMPETENT	COMPETENT	YES	NO/YES
10	M	14	20	CPO	COMPETENT	EMERGING	YES	NO/YES
11	F	12	17	BCLP	COMPETENT	COMPETENT	YES	YES/YES
12	M	14	20	UCLP	COMPETENT	COMPETENT	YES	NO/YES
13	F	13	23	BCLP	COMPETENT	COMPETENT	YES	NO/YES
14	M	14	22	BCLP	COMPETENT	EMERGING	YES	NO/NO
15	M	12	18	BCLP	EMERGING	EMERGING	YES	YES/NO
16	M	-	18	CL/SUBM	COMPETENT	EMERGING	NO	NO/NO
17	M	12	23	UCL/BCP	COMPETENT	COMPETENT	YES	YES/NO
18	M	12	17	UCLP	COMPETENT	COMPETENT	UNKNOWN	NO/NO
19	M	14	24	UCLP	EMERGING	EMERGING	YES	NO/NO
20	M	14	24	UCLP	EMERGING	EMERGING	YES	YES/YES (one ear)
21	F	14	19	CPO	COMPETENT	-	YES	YES/NO
22	M	13	20	BCLP	EMERGING	-	YES	UNKNOWN /YES

Procedures

Prior to the first recording, all caregivers reported their child's expressive vocabulary by completing the Danish version of the MacArthur-Bates Communicative Developmental Inventories (CDI: Andersen, Vach, Wehberg, & Bleses, 2006; Fenson et al., 2007). The children were video recorded for 2 × 45 minutes on two separate days in the clinic during natural play with a caregiver. Available toys/objects represented words used most frequently by Danish toddlers (Wehberg et al., 2007) as well as all Danish consonant phonemes. The video recordings were separated into files of approximately 22 minutes each to account for memory constraints. RTL was used for assessment of consonant inventory, and three raters were trained in the RTL procedure. Two raters were SLPs with 10–12 years' experience with children with CP, and one rater was a graduate student with previous experience in rating speech in children with CP. The raters listened to an entire recording in real time without pausing. No notes were taken. Raters pressed a key every time they heard a syllable. Syllables were annotated in a software program developed in an ongoing study, Timing Of Primary Surgery for cleft palate (TOPS) (Appelbe & Willadsen, 2017). Syllables were defined as a speech-like vocalisation including at least one vowel, and reflexive and whispered vocalisations were not counted. After listening to an entire recording, raters wrote down consonants and syllable shapes they felt the child commanded. The mean number of syllables evaluated per child was 530 syllables (min-max: 231–662, median: 543 syllables). Because the children produced very different amounts of syllables within the given time frame, some children were evaluated based on one recording while others were evaluated based on all four recordings. Intra-rater reliability was substantial to almost perfect for consonant inventories, κ = 0.801, 0.795, and 0.886, respectively.

Finally, each video recording was viewed independently without pause by two SLPs with 2–10 years of experience working in cleft clinics who were unfamiliar with the participants. Based on their clinical opinion, the SLPs were asked whether or not a child needed intervention, and whether this recommendation was based on consonant production, expressive vocabulary, receptive language, social interaction skills, or other reasons. The SLPs were informed of each child's age, but had no knowledge of the purpose of this study.

Statistical analyses

Kappa and intraclass correlation (ICC) values in this article are interpreted based on commonly cited benchmarks: Kappa: <0 no agreement, 0–.20 slight, .21–.40 fair, .41–.60 moderate, .61–.80 substantial and .81–1 almost perfect (Landis & Koch, 1977); ICC: <.40 poor, .40–.59 fair, .60–.74 good and .75–1.0 excellent (Cicchetti, 1994). Statistical analyses were performed using SPSS statistical package version 22 (SPSS Inc., Chicago, IL).

The screening procedure

The screening procedure consisted of two steps: First, consonant inventories were established by RTL assessment. Second, predictive measures from the consonant inventories were used to assign children to +/− need for intervention conditions. Predictive measures of later speech performance were used for the screening procedure, i.e. true consonant inventory, oral consonant inventory, frequency and number of oral consonants, emerging consonants, frequency of oral stops, number of dental stops, and observed expressive vocabulary (see table 1). The RTL procedure does not allow assessment of absolute numbers and frequencies of consonants produced. Instead, children were assigned to the 'need for intervention' condition based on absence of the predictive measures: true consonants, oral consonants, oral stops, and/or dental stops. If a low number of these consonant types predicts lower PCC-A scores at a later age, then surely absence of these consonants must also predict low PCC-A scores. Nevertheless, in reducing consonant inventory, frequency, and total counts to absence/presence there is a risk of measures being too crude for a sufficiently sensitive screening procedure. Emerging consonants were excluded as a predictive measure since the definition of an emerging consonant entails evaluation of correctness which again is not possible within the frame of RTL. Also, RTL is not designed to assess vocabulary. One study of children with CP (Scherer & D'Antonio, 1995) and several studies of children without CP (see Fenson et al., 1994) have found substantial correlation between reported and observed vocabulary. Therefore, observed expressive vocabulary was replaced by parent-reported expressive vocabulary, assessed by the CDI. The predictive value of the CDI has not been systematically evaluated in children with CP older than 12 months; yet, expressive vocabulary is the best predictor of later language skills in children without CP (Rescorla, 2011). However, reported expressive vocabulary as a predictor of later speech-language skills has been shown to have high sensitivity, but low specificity in this age group (Henrichs et al., 2011; Westerlund, Berglund, & Eriksson, 2006). Cut-off score for language delay is typically set at the 10th percentile on the CDI (Henrichs et al., 2011) and was also chosen for this study. The predictive measure 'dental stops' was found in Swedish-learning children, but Danish does not have dental stops. Still, Swedish and Danish have highly similar phonological systems: both languages have two contrastive sets of stops, /p t k/ and /b d g/, the main difference being that /b d g/ are voiced in Swedish but unvoiced in Danish. In Swedish, /t/ is dental while /d/ can be dental or alveolar, whereas in Danish, /t d/ are both alveolar, and /t/ is mostly affricated (Basbøll, 2005; Riad, 2014). Based on the similarities of the phonological systems, absence of alveolar stops was used as a predictive measure in the screening procedure. It is likely that a combination of predictive measures will yield a higher predictive value than a single predictive measure. However, this has not been investigated. Therefore, a child was assigned to the 'need for intervention' condition if at least one of the predictive measures was absent according to the consonant inventories assessed by the RTL procedure, or if the CDI score was at or below the 10th percentile.

Results

Consonant measures from RTL assessment

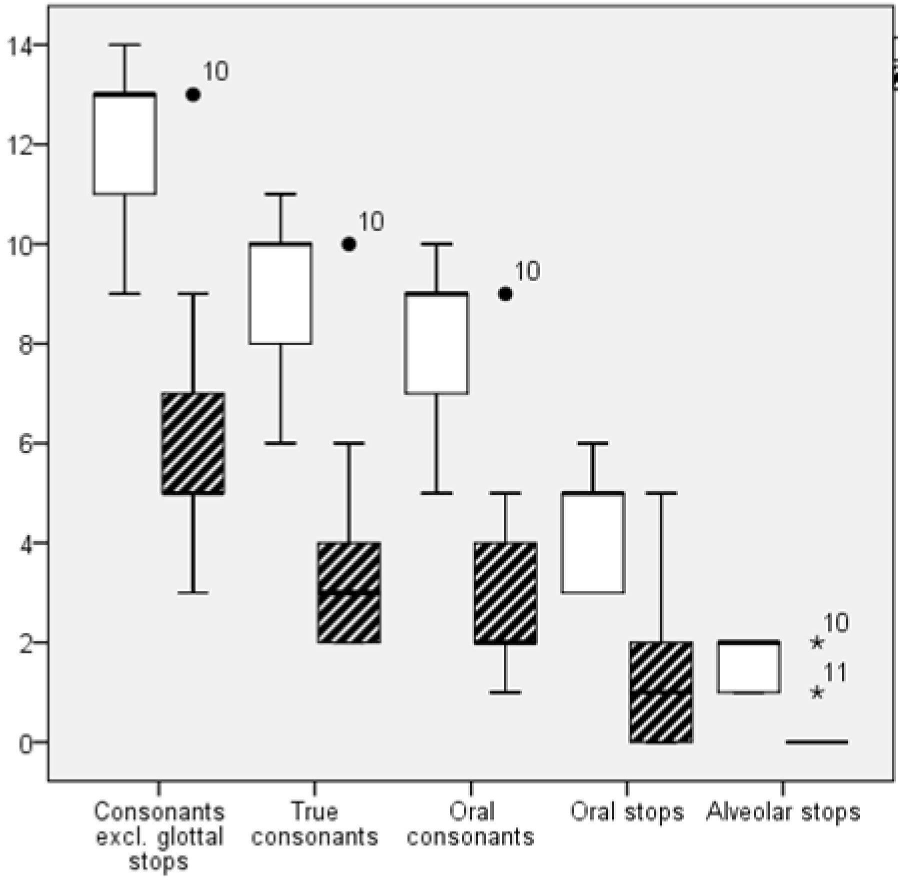
Consonant inventories established from the RTL assessment are shown in appendix 1 for all children. ICC estimates and their 95% confidence intervals were calculated based on a mean-rating (k = 3), absolute-agreement, two-way random-effects model. Absolute agreement was chosen to account for systematic variability between raters. The results for both average measures and single measures are reported as the purpose of the reliability testing is two-fold: to investigate if RTL assessment of consonant inventory is reliable across three raters and for single raters. All consonant measures (inventory and no. of different consonants excluding glottal stops, inventory and no. of different true consonants, inventory of oral consonants, and inventory of different oral stops) showed excellent reliability for average measures and good to excellent agreement for single measures (see table 3). All results were significant at the $p < .001$-level.

Table 3. Agreement on consonant measures from RTL assessment.

Consonant measure	Form	ICC	p-value	95% CI
Consonant inventory excl. glottal stops	Average measures	.897	p<0.001	[0.88-0.91]
	Single measures	.743	p<0.001	[0.70-0.78]
No. of different consonants excl. glottal stops	Average measures	.946	p<0.001	[0.89-0.98]
	Single measures	.853	p<0.001	[0.72-0.93]
True consonant inventory	Average measures	.885	p<0.001	[0.86-0.91]
	Single measures	.719	p<0.001	[0.67-0.77]
No. of different true consonants	Average measures	.945	p<0.001	[0.88-0.98]
	Single measures	.852	p<0.001	[0.71-0.93]
Oral consonant inventory	Average measures	.902	p<0.001	[0.88-0.92]
	Single measures	.755	p<0.001	[0.71-0.80]
No. of different oral consonants	Average measures	.961	p<0.001	[0.92-0.98]
	Single measures	.890	p<0.001	[0.78-0.95]
Oral stop inventory	Average measures	.891	p<0.001	[0.86-0.92]
	Single measures	.732	p<0.001	[0.66-0.79]
No. of different oral stops	Average measures	.969	p<0.001	[0.94-0.99]
	Single measures	.913	p<0.001	[0.83-0.96]

Raters agreed 100% that all children produced oral consonants and true consonants. Fleiss' kappa was performed to determine inter-rater reliability for absence/presence of oral stops and alveolar stops. For oral stops, the inter-rater reliability was almost perfect, $\kappa = 0.847$ (disagreement in two cases), and for alveolar stops, intra-rater reliability was substantial, $\kappa = 0.758$ (disagreement in four cases). Disagreement was solved by majority rule. As shown in table 4, oral stops were absent in six children (27%), alveolar stops were absent in nine children (41%), and eight children (36%) had a CDI score at or below the 10th percentile. In total, 13 children (59%) matched one or more of the 'need for intervention' criteria. Figure 1 shows how many different consonants, true consonants, oral consonants, oral stops, and alveolar stops the children produced in the 'no need for intervention' group compared to the 'need for intervention' group.

Figure 1. Consonant inventories (agreed by at least two raters) form the RTL procedure in the 'need for intervention' (white) vs. the 'no need for intervention' (striped) group.



Agreement between SLPs' evaluations

Cohen's kappa was calculated to determine inter-rater reliability between the SLPs evaluation of need for intervention and between the SLPs' rationales for recommending intervention (consonant production and expressive vocabulary). Reliability for the rationales of receptive language, social interaction skills and other reasons were not considered since they were only used by SLPs a few times, and they were not a part of the screening procedure. Almost perfect reliability was found between SLPs on which children needed intervention (κ = 0.908) (disagreement in one case), though observation was recommended by one SLP in two cases. Almost perfect reliability was also found between SLPs' recommendation of intervention based on both consonant production (κ = 0.820, disagreement in two cases) and expressive vocabulary (κ = 0.891, disagreement in one case).

Agreement between SLPs' evaluations and the screening procedure

Cohen's kappa was also calculated to determine inter-rater reliability between SLPs' evaluation of need for intervention and the screening procedure and between the SLPs' rationales for recommending intervention (consonant production and expressive vocabulary) and the inclusion criteria of the screening procedure. Inter-rater agreement between the SLPs and the screening procedure was almost perfect (κ = 0.816); there was disagreement between SLPs in one case, and in one other case, the SLPs found no need for intervention while the child met the inclusion criteria of the screening procedure. Reliability was substantial for SLPs' recommendation of intervention based on consonant production and the consonant measures of the screening procedure (true and oral consonants, oral and dental stops) (κ = 0.727, disagreement in three cases), and moderate to substantial for SLPs' recommendation of intervention based on expressive vocabulary and the CDI criterion of the screening procedure (κ = 0.585–0.645, disagreement in four cases).

Table 4 shows absence/presence of oral stops and alveolar stops as well as which children met the inclusion criteria for the 'need for intervention' condition based on both the screening procedure and the evaluation by SLPs. Since all children had oral and true consonants in their inventories, these categories are not shown here. Specificity, sensitivity, and positive and negative predictive values are shown in table 5.

Table 4. Results from the screening procedure and the SLPs' evaluation of need for intervention.

Predictive measure	Child /rater	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22
Oral stops	1	P	P	P	P	P	P	P	P	P	P	P	P	P	P	P	P	*A*	*A*	*A*	*A*	*A*	*P*
	2	P	P	P	P	P	P	P	P	P	P	P	P	A	P	P	P	*A*	*A*	*A*	*A*	*A*	*A*
	3	P	P	P	P	P	P	P	P	P	P	P	P	P	P	P	P	*A*	*A*	*A*	*A*	*A*	*A*
Alveolar stops	1	P	P	P	P	P	P	P	P	P	P	P	*P*	*A*	*A*	*P*	*A*	*A*	*A*	*A*	*A*	*A*	*A*
	2	P	P	P	A	A	P	P	P	P	P	P	*A*	*A*	*A*	*A*	*A*	*A*	*A*	*A*	*A*	*A*	*A*
	3	P	P	P	P	P	P	P	P	P	P	P	*A*	*A*	*A*	*A*	*A*	*A*	*A*	*A*	*A*	*A*	*A*
CDI score </=10^{th} perc.		N	N	N	N	N	N	N	N	N	*Y*	*Y*	N	N	N	*Y*	*Y*	N	N	*Y*	*Y*	*Y*	*Y*
Int. need screening		N	N	N	N	N	N	N	N	N	*Y*	*Y*	*Y*	*Y*	*Y*	*Y*	*Y*	*Y*	*Y*	*Y*	*Y*	*Y*	*Y*
Int. need SLPs	1	N	N	N	N	N	N	N	N	N	N	*Y*	N	*Y*	*Y*	*Y*	*Y*	*Y*	*Y*	*Y*	*Y*	*Y*	*Y*
	2	N	N	N	N	N	N	N	N	N	*Y*	*Y*	N	*Y*	*Y*	*Y*	*Y*	*Y*	*Y*	*Y*	*Y*	*Y*	*Y*

P = present, A = absent, N = no, Y = yes. Italics indicate need for intervention.

Table 5. Specificity and sensitivity of the screening procedure compared to SLPs' evaluation.

SLPs/screening	Need for intervention	No need for intervention	
Need for intervention	11 (12)	0	Sensitivity = 1.00, 95% CI [0.72-1.00] (Sensitivity = 1.00, 95% CI [0.74-1.00])
No need for intervention	2 (1)	9	Specificity = 0.81, 95% CI [0.48-0.98] (Specificity = 0.90, 95% CI [0.56-1.00])
	PPV = 0.85, 95% CI [0.61-0.95] (PPV = 0.92, 95% CI [0.65-0.99])	NPV = 1.00	

PPV = positive predictive value, NPV = negative predictive value.

Discussion

RTL assessment

The first purpose of this study was to investigate if trained raters can reliably assess consonant inventory, including predictive measures of later speech-language performance, in children with CP using an RTL procedure. Excellent agreement across three raters on measures of consonant inventory suggests that RTL is a reliable way of screening the consonant inventory of toddlers with CP. No. of different oral consonants and no. of different oral stops also reached excellent agreement for single measures, whereas the other consonant measures had broader confidence intervals resulting in good to excellent agreement. Thus, although RTL is a reliable method for assessing consonant inventory in toddlers with CP across raters, single-rater assessment might not be as reliable. Still, the results are encouraging compared to inter and intra-rater percentage agreement for phonetic transcription of children's speech ranging from 60–80% (Shriberg & Lof, 1991).

Raters agreed 100% on absence/presence of oral consonants and true consonants. However, since all children produced both oral and true consonants, absence or presence of these categories is not a good candidate for a screening procedure. Almost perfect agreement on presence or absence of oral stops and substantial agreement on presence or absence of alveolar stops suggest that RTL is a reliable way of screening predictive measures of later speech-language skills in toddlers with CP.

Agreement between the screening procedure and SLPs' clinical judgement

The second question posed in this study was if experienced SLPs' clinical judgement agrees with the screening procedure as to which children need intervention. The SLPs' judgement was in almost perfect agreement with the screening procedure (20 out of 22 cases, $\kappa = 0.816$), suggesting that the screening procedure has high external validity. The SLPs recommended intervention in 11 cases (disagreement in one case). These children also needed intervention according to the screening procedure, indicating high sensitivity. The SLPs found that 10 children did not need intervention (disagreement in one case) which was in disagreement with the screening procedure in two cases, also indicating high specificity (see table 5). Albeit levels of sensitivity and specificity are notable, confidence intervals are very broad due to the small sample size. However, the screening procedure found that 59% (13 out of 22) of the children with CP needed intervention. This finding is in accordance with previously reported prevalence of speech delay in 3-year-olds with CP (Lohmander, 2011), and is also similar to no. of children with CP referred for early speech-language intervention in studies by Hardin-Jones and Chapman (2008) and Hardin-Jones and Jones (2005). Therefore, it is reasonable to assume that the screening procedure can actually discriminate between children with CP who need intervention and those who do not. The SLPs' agreement on the rationale for recommending intervention was almost perfect for both consonant production (κ = 0.820) and expressive vocabulary (κ = 0.891). Agreement between SLPs' recommendation of intervention based on consonant production and the screening procedure's consonant measures was also substantial (κ = 0.727) while agreement between SLPs' recommendation of intervention based on expressive vocabulary and the screening procedure's CDI criterion was moderate to substantial (κ = 0.585–0.645). This implies that although the overall sensitivity and specificity of the screening procedure is high, the validity of the single predictive measures might not be as high. In the following, disagreements between raters, the screening procedure, and SLPs will be explored in a qualitative manner.

Disagreements on need for intervention based on consonant measures

The SLPs found that all six children without oral stops needed intervention, and in five cases, heir recommendation was based on consonant production (disagreement in one case, child 19). Furthermore, one SLP noted in five out of six cases that children did not produce any or very few obstruent sounds. The SLPs recommended that 11 of 12 children with absent alveolar stops needed intervention. With the exception of one child (child 12) and the abovementioned case of disagreement, SLPs recommendation of intervention was based on consonant production. In the RTL assessment, one rater found that child 12 mastered one alveolar stop whereas the other raters both made a note saying they might have heard an alveolar stop, but weren't sure. Since the aim of RTL listening is to estimate what the child is in control of, there is a risk of low frequency or emerging sounds in a child's repertoire being missed in this screening procedure. This might explain the latter disagreement between SLPs and the screening procedure. Another explanation might be that child 12 only had absence of alveolar stops, but other oral stops present and a CDI score above the 10th percentile. Yet, two other children in this study had the same profile (child 13 and 14). The SLPs agreed that these two children needed intervention. Since child 13 and 14 had rather large vocabularies (well above 100 words) while child 12 had less than 50 words, it could be speculated that phonological patterns, that are more likely to emerge when a child's vocabulary exceeds 50 words (Stoel-Gammon, 2011), played a role in the SLPs' judgement. Indeed, child 13 and child 14 showed patterns of backing and glottal compensatory articulation. A third explanation might be that absent alveolar stops is not a good predictive measure. Although the results showed substantial agreement between raters in judging absence/presence of alveolar stops (κ = 0.758), it was still the lowest agreement of the predictive measures in the screening procedure. Furthermore, albeit Klintö et al. (2014) found that no. of dental stops correlated with PCC-A score at 3 years, absolute numbers are highly sensitive to sample size. When Klintö et al. (2014) excluded the two least voluble children in the study, no significant correlation was found. Willadsen (2007) found that frequency of alveolars at 11 months of age was a strong predictor of PCC at 3 years, but found no correlation between frequency of alveolars at 18 months of age and PCC at 3 years, however; Willadsen (2007) did not report specifically on alveolar stops, but only on alveolar place. Lastly, a difference between languages may also exist. De Boysson-Bardies and Vihman (1991) found that Swedish-learning children (aged 9–17 months) produced a high percentage of dental stops compared to children learning French, Japanese and English which might imply that producing few dental stops have a relative greater impact on later speech production skills in Swedish-learning children.

Disagreements on need for intervention based on expressive vocabulary size
In the present study, the SLPs had no knowledge of the children's CDI scores. SLPs found expressive vocabulary to be within normal limits in four cases where the CDI indicated a score at or below the 10th percentile, and in two other cases, CDI percentile scores were normal while the SLPs found need for intervention based on expressive vocabulary (disagreement between SLPs in one case, child 10). However, intervention was recommended for all but one child (child 10) with a CDI score at or below the 10th percentile. In this case, SLPs disagreed on whether or not the child needed intervention. Child 10 had a low CDI score, but matched none of the other predictive measures. One other child (child 11) had the same profile, but while child 10 scored at the 10th percentile, child 11 scored at percentile 2.5, indicating that a score at the 10th percentile is not in itself a good predictor, but must be combined with other predictive measures. Interestingly, child 10 had almost twice as many different consonants, oral consonants and true consonants as child 11 (see appendix 1 and figure 1 in which child 10 appears to be an outlier in the 'need for intervention' group) which might imply that these consonant measures can be relevant predictive measures in combination with CDI score. In addition, consideration should be given to changing the CDI criterion to a lower percentile in cases where a low CDI percentile is the only risk indicator.

Limitations

Although the raters participating in this study had good inter-rater reliability scores, it cannot be assumed that these scores will apply to other raters. Hence, further evaluation of the screening procedure is needed to confirm its applicability to research and clinical practice. Also, this study only included 22 children with CP and should be replicated with more children and more raters.

Albeit, only predictive measures obtained after surgery were used in this screening procedure, it is possible that predictive measures obtained in the study by Klintö et al. (2014) in which children had received two-stage palatal closure only apply to children receiving this surgical procedure. This might especially concern alveolar stops. Also, all predictive measures used in this study were found in studies of children with unilateral cleft lip and palate while the children in this study had different cleft types. Conducting a larger study with the possibility of evaluating the screening procedure in children with different cleft types is recommended. Finally, the selected predictive measures for the screening procedure are based on studies of two different languages; English and Swedish, while all children in this study were acquiring Danish. It is possible that the different phonological systems affect which predictive measures are more relevant.

Conclusion

Consonant inventory including suggested early predictive measures of later speech and language performance can be reliably assessed by trained listeners with an RTL procedure and the CDI. Almost perfect agreement between the clinical opinion of experienced SLPs and the screening procedure in judging which children need intervention indicates that the screening procedure is a valid procedure for identifying children with CP who need early intervention. Absent oral stops, and possibly absent alveolar stops in combination with a CDI score of expressive vocabulary below the 10th percentile seem to be good candidates for a screening procedure.

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Declaration of interest

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Appendix 1. Consonant inventories by rater and inventory agreed by at least two raters (agreed inventory) for all children

Child	Rater	Consonant inventory	Agreed inventory	Child	Rater	Consonant inventory	Agreed inventory
1	1 2 3	b̥ d̥ g̊ p^{h} t^{s} k^{h} s w j m n ŋ h ʔ b̥ d̥ p^{h} t^{s} k^{h} f s w j m n ŋ h b̥ d̥ g̊ p^{h} t^{s} k^{h} s w j m n ŋ h	b̥ d̥ g̊ p^{h} t^{s} k^{h} s w j m n ŋ h	12	1 2 3	b̥ d̥ g̊ w l j m n ŋ h b̥ g̊ w j m n h b̥ g̊ p^{h} w j m n h	b̥ g̊ w j m n h
2	1 2 3	b̥ d̥ g̊ w j m n ŋ h b̥ d̥ g̊ w j m n h b̥ d̥ g̊ w j m n ŋ h	b̥ d̥ g̊ w j m n ŋ h	13	1 2 3	b̥ g̊ s j m n ŋ h ʔ w j m h g̊ w j m n ŋ h ʔ	g̊ w j m n ŋ h ʔ
3	1 2 3	b̥ d̥ g̊ t^{s} k^{h} s w j m n h b̥ d̥ g̊ p^{h} t^{s} f s w j m n h b̥ d̥ g̊ p^{h} t^{s} k^{h} f s w j ʁ m n ŋ	b̥ d̥ g̊ p^{h} t^{s} k^{h} f s w j m n h	14	1 2 3	b̥ g̊ s w j m n ŋ h b̥ g̊ w j m n h b̥ g̊ k^{h} s w j m n ŋ h	b̥ g̊ s w j m n ŋ h
4	1 2 3	b̥ d̥ g̊ p^{h} t^{s} k^{h} s w l j m n ŋ h b̥ p^{h} k^{h} w l j m n ŋ h b̥ d̥ g̊ p^{h} k^{h} s w l j m n ŋ h	b̥ d̥ g̊ p^{h} k^{h} s w l j m n ŋ h	15	1 2 3	b̥ d̥ l j m h ʔ b̥ w j m h b̥ l j m n h	b̥ l j m h
5	1 2 3	b̥ t^{s} l j n b̥ g̊ p^{h} w j m n h b̥ d̥ p^{h} t^{s} s w l j m n h	b̥ p^{h} t^{s} w l j m n h	16	1 2 3	b̥ w j m h b̥ m h b̥ m h	b̥ m h
6	1 2 3	b̥ d̥ g̊ t^{s} k^{h} f s w l j m n h b̥ d̥ g̊ t^{s} k^{h} f s w l j m n ŋ h b̥ d̥ g̊ p^{h} t^{s} k^{h} f s w l j ʁ m n ŋ h	b̥ d̥ g̊ t^{s} k^{h} f s w l j m n ŋ h	17	1 2 3	w j m ŋ h ʔ w j m n ŋ h w j m n ŋ h ʔ	w j m n ŋ h ʔ
7	1 2 3	b̥ d̥ g̊ t^{s} k^{h} f s w j m ŋ h b̥ d̥ g̊ t^{s} f w j m n h b̥ d̥ g̊ p^{h} t^{s} k^{h} f s w l j m n ŋ h	b̥ d̥ g̊ t^{s} k^{h} f s w j m n ŋ h	18	1 2 3	w j m ŋ h ʔ j m n ŋ h w j m ŋ h	w j m ŋ h
8	1 2 3	b̥ d̥ g̊ t^{s} f s w l j m n h b̥ d̥ p^{h} t^{s} f s w l j m n h b̥ d̥ t^{s} f s w l j m n h	b̥ d̥ t^{s} f s w l j m n h	19	1 2 3	l j m n ŋ h w j m n h j m n h	j m n h
9	1 2 3	b̥ d̥ g̊ k^{h} s w j m n ŋ h b̥ d̥ g̊ p^{h} k^{h} s w j m n h b̥ d̥ g̊ k^{h} s w j m n ŋ h	b̥ d̥ g̊ k^{h} f s w j m n ŋ h	20	1 2 3	w j m n h ʔ w j m n h w j m n h	w j m n h
10	1 2 3	b̥ d̥ g̊ p^{h} k^{h} f s w j m n ŋ h b̥ d̥ g̊ t^{s} f s w j m n ŋ h b̥ d̥ g̊ t^{s} k^{h} f s w l j m n ŋ h	b̥ d̥ g̊ t^{s} k^{h} f s w j m n ŋ h	21	1 2 3	w j m n ŋ h ʔ w j m n h w j m n h	w j m n h
11	1 2 3	d̥ g̊ w j m n ŋ h d̥ g̊ w j m n h d̥ g̊ w j m n	d̥ g̊ w j m n h	22	1 2 3	b̥ w j m n ŋ h ʔ w j m n h w j m n h ʔ	w j m n h ʔ

STUDY 2

Early intervention in toddlers with cleft palate
– Short-term effects of a parent-implemented focused stimulation program

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Abstract

Purpose

This study investigated the short-term effects of a parent-implemented focused stimulation intervention targeting productive vocabulary and consonant inventory directly in toddlers with cleft palate ± cleft lip (CP) and delayed speech and/or language development.

Method

A quasi-experimental pre-test post-test control group design was used to compare outcomes from 14 toddlers with delayed speech and/or language development and CP who received intervention (INT group) to a pairwise matched control group of 14 toddlers with CP (CONT group). Only INT parents received nine home-based sessions with instruction in focused stimulation strategies.

Results

The INT group had significantly higher gain scores with large effect sizes for lexical age, a measure of observed vocabulary, and for true consonant types in words at post-test than the CONT group. The INT group were eight times more likely to not need intervention than the CONT group at post-test. All parents in the INT group reported positive effect on vocabulary and interaction skills. In general, parents were satisfied with the intervention program.

Conclusions

Parent-implemented focused stimulation had positive short-term effects on productive vocabulary and consonant inventory in toddlers with CP and delayed speech and/or language development when compared to a matched control group. Long-term results will be collected and reported in a separate study. Most productive vocabulary and consonant inventory measures increased whether or not the children received intervention which underlines the importance of including matched control groups in intervention studies of toddlers with CP.

Introduction

In typical development, speech sound development is closely linked to productive vocabulary development (Stoel-Gammon, 2011). There are substantial overlaps between prelinguistic sounds and syllable shapes and early words. Children choose words beginning with consonants that are already part of their prelinguistic phonetic inventory, henceforth termed consonant inventory to exclude vowels, and avoid words with initial consonants outside their inventory: a phenomenon known as lexical selectivity (Ferguson & Farwell, 1975; Leonard, Schwartz, Morris, & Chapman, 1981; Vihman, Macken, Miller, Simmons, & Miller, 1985). When babies repeat the same CV-syllable over and over again, the practice and eventual mastery of this syllable help create an essential link between articulatory movement and the resulting acoustic signal: the auditory-articulatory feedback loop (Fry, 1966; Stoel-Gammon, 1998a). Well-practiced canonical syllables may act as 'building blocks' for early words (Ramsdell, Oller, Buder, Ethington, & Chorna, 2012; Vihman et al., 1985). Once a small productive vocabulary is established, children will typically generalise one or more of their production patterns into a phonological template where words are holistic matches of well-practised building blocks (Velleman & Vihman, 2002). As children expand their vocabularies towards 100 words, it is assumed that they begin to recognise similarities across phonological forms (Walley, Metsala, & Garlock, 2003; Stoel-Gammon, 1998a). Information about phonological forms is organised as underlying phonological representations to distinguish words in the expanding vocabulary (Beckman & Edwards, 2000). This, in turn, leads to a more systematic phonology in which more consistent phonological processes can be observed (Ferguson & Farwell, 1975; Vihman, 2014).

Interactions between vocabulary and consonant inventory in toddlers with CP

The link between speech sound development and productive vocabulary development can also be observed in toddlers with CP. Prior to palatal surgery, babies with cleft palate ± cleft lip (CP) have difficulties with sound production, especially oral pressure consonants. After surgery, many toddlers with CP show persistent delays in consonant acquisition. As a group, toddlers with CP post-surgery have smaller consonant inventories. This finding is reported in the majority of studies comparing the speech of toddlers with CP and complete palate repair to their peers without CP before age two (Chapman, Hardin-Jones, & Halter, 2003; Salas-Provance, Kuehn, & Marsh, 2003; Willadsen, 2012). In addition, consonant inventories of toddlers with CP are characterised by fewer oral stops (Chapman et al., 2003; Jones et al., 2003; Salas-Provance et al., 2003; Willadsen, 2012), fewer true consonants (Chapman et al., 2003), fewer alveolar consonants (Jones et al., 2003; Willadsen, 2012), more nasals and glides (Jones et al., 2003), and more velars and glottals (Willadsen, 2012). At the same time, studies have reported slower productive vocabulary development from ages 14-30 months compared to peers without CP (Broen, Devers, Doyle, Prouty, & Moller, 1998; Hardin-Jones & Chapman, 2014; Jocelyn, Penko, & Rode, 1996; Lu, Ma, Luo, & Fletcher, 2010; Morris, 1962; Neiman & Savage, 1997; Scherer & D'Antonio, 1995). Therefore, it seems that the restricted consonant inventories of toddlers with CP provide them with fewer building blocks for producing early words. Furthermore, their first words are more likely to begin with nasals, approximants, and vowels compared to peers without CP, and more labials, velars, and glottals (Estrem & Broen, 1989; Hardin-Jones & Chapman, 2014; Willadsen, 2013), thus reflecting lexical selectivity. This indicates that toddlers with CP have fewer words to select from to create their early productive vocabularies. In particular, the low frequency of stops in early productive vocabulary is concerning because almost half of typically developing Danish toddlers' first 50 words (Wehberg et al., 2007) begin with a stop consonant. In addition, percentage of oral stops and dental stop tokens at 17-18 months correlate with measures of consonant accuracy at 27 and 36 months (Hardin-Jones & Chapman, 2008; Klintö et al., 2014).

Phonological development in children with CP is affected by early phonetic constraints. Harding and Grunwell (1996) proposed that consonant inventories that were initially shaped by the structural deficit may over time affect phonological development as toddlers develop phonological rules from words build from restricted consonant inventories. Studies have reported lower PCC scores in 2 and 3-year-olds with CP (e.g. Chapman, 1993; Chapman & Hardin; 1992; Klintö et al., 2014; Willadsen, 2012) and use of more phonological processes at age 3 compared to peers without CP (Chapman, 1993; Klintö et al., 2014). In addition, prerequisites for reading may also be affected and related to speech performance (Chapman, 2011).

Targeting vocabulary and consonant inventory in early intervention

Early intervention studies in toddlers with speech-language delays are generally rooted in social-interactionist theory and have therefore primarily focused on expanding productive vocabulary and other language measures rather than consonant inventory or phonology (e.g. Buschmann et al., 2008; Gibbard, Coglan, & MacDonald, 2004). Within the social-interactionist paradigm, language learning is believed to occur in meaningful interactions, implying that language forms should never be separated from language meaning (Fey, 1986). Very few studies have explored the possible indirect effect on consonant inventory and phonology of targeting productive vocabulary in toddlers (Girolametto, Pearce, & Weitzman, 1997; Whitehurst et al., 1991). Whitehurst et al. (1991) employed parent-implemented enhanced milieu teaching (EMT; Kaiser, Hancock, & Hester, 1998) in toddlers aged 24-36 months with large gaps between receptive and expressive language. Target words included sounds within participants' pre-test consonant inventories. When compared to a matched non-treatment control group, intervention effect was found for productive vocabulary, but not for percent consonants correct (PCC: Shriberg & Kwiatkowski, 1982). This led these researchers to recommend separate targeting of phonology at a later time point. Expanding on this finding, Girolametto et al. (1997) reported that parent-implemented focused stimulation (FS) affected consonant inventory indirectly in toddlers aged 23-33 months with small vocabularies for age compared to a matched non-treatment control group. Both productive vocabulary and consonant inventory expanded as a function of intervention, but PCC did not. Girolametto et al. (1997) noted that these results mirror the simultaneous expansion of vocabulary and consonant inventory in typically developing toddlers (see Stoel-Gammon, 2011) and proposed that the observed vocabulary expansion required more contrasting phoneme categories for word organisation, in turn leading to greater variation in consonant inventory but not necessarily accuracy.

Scherer (1999) built on Girolametto et al.'s (1997) findings in the first experimental study of early intervention in language-delayed toddlers with CP. Scherer (1999) investigated the effects of EMT in a multiple baseline design including three toddlers with CP aged 24-28 months with reported productive vocabularies between 25-84 words. However, whereas Girolametto et al. (1997) targeted consonant inventory and phonology indirectly by using target words with consonants within the participants' pre-test consonant inventories, Scherer (1999) included target words with consonants both within and outside the participants' pre-test consonant inventories thus targeting both vocabulary and consonant inventory directly. Scherer (1999) found that participants learned most words readily. This indicates that including word targets beginning with consonants outside participants pre-test inventory does not prevent acquisition of these word targets. Scherer (1999) also reported changes in productive vocabulary and consonant inventory from pre-test to post-test; however, only target word use were monitored at multiple time points, but none of the outcome measures. Thus, it cannot be determined whether the observed changes in productive vocabulary and consonant inventory were due to intervention or general development (including both maturation and learning).

Scherer, D'Antonio and McGahey (2008) also targeted both vocabulary and consonant inventory directly by including stop consonants in target words. It is presumed that participants did not produce these stops at pre-test. Parents were trained to use FS procedures with ten toddlers with CP aged 18-35 months. Seven of these toddlers had reported productive vocabularies between 1-59 words whereas three toddlers had larger reported productive vocabularies between 123-324 words. Scherer et al. (2008) reported increase in productive vocabulary and consonant inventory; however, similar gains were observed in the control group of language-matched toddlers without CP. Hence, this study did not show intervention effect beyond general development. Scherer et al. (2008) acknowledged the limitations of a language-matched control group and concluded that intervention did not result in the intervention group obtaining typical development. Despite the limitations in study design, Scherer et al. (2008) reported another important outcome, namely that parents were able to learn language support strategies.

In a recent study, Kaiser, Scherer, Frey, & Roberts (2017) reported results from a small randomised controlled trial of 19 toddlers with CP aged 15-36 months where vocabulary and consonant inventory were also targeted directly. Eight toddlers received EMT while 11 toddlers received business-as-usual treatment. Kaiser et al. (2017) found significant group differences in reported productive vocabulary gain and PPC-A gain from pre-test to post-test. However, participants in the intervention group had considerably lower scores at pre-test than the control group (reported vocabulary mean: 182.0 vs. 303.9; PCC-A mean: 31.8 vs. 45.7). Hence, in spite of random group allocation, groups were not equivalent at pre-test and the reported effects on vocabulary and PCC-A in should thus be interpreted with caution. Furthermore, the participants in Kaiser et al. (2017) had much larger vocabularies than participants in the studies by Scherer and colleagues (1999; 2008) as determined by MacArthur-Bates Communicative Developmental Inventories (CDI: Fenson et al., 2007) and were probably not language-delayed. Information on mean age and reported vocabulary size indicate average CDI percentile scores well within the normal range (Dale & Fenson, 1996).

Two more experimental studies of early intervention in toddlers with CP exist, but they do not target vocabulary and consonant inventory directly (Broen, Doyle, & Bacon, 1993; Ha, 2015).

As general development has not been appropriately controlled for in the above-mentioned studies, and control groups have not been sufficiently matched, there is still no compelling evidence of short-term effects of targeting vocabulary and consonant inventory directly in early intervention in toddlers with CP, and long-term effects have not been reported.

Speech-language inclusion criteria

Not all toddlers with CP are speech-language delayed (Chapman & Willadsen, 2011; Hardin-Jones & Chapman, 2008). Speech-language inclusion criteria are thus necessary to identify toddlers that are at risk of later speech-language difficulties. In addition, it will ensure representativeness of participants and provide a foundation for matching of control groups. Identification must be time-efficient while still representing a reliable and valid evaluation of speech and language performance. This is a particular challenge with toddlers since speech norms for children younger than three years are non-existent, and available language tests are difficult to undertake in toddlers and may not detect less severe language delays (Scherer, Frey, & Kaiser, 2014). Jørgensen and Willadsen (2017) aimed to construct a screening procedure based on predictive measures of later speech-language difficulties in toddlers with CP (see table 1). These measures were derived from longitudinal studies of toddlers with completely repaired CP (Chapman, 2004; Klintö et al., 2014; Hardin-Jones & Chapman, 2008).

Table 1. Predictive measures at 17-21 months correlating with later speech-language difficulties.

17-21 months	27 months	36-39 months	No. of children	Reference
True consonant types		NDW	15	Chapman, 2004
Oral consonant types		PCC-A	9	Klintö et al., 2014
Oral consonant tokens		PCC-A	9	Klintö et al., 2014
Emerging consonant tokens		emerging consonant tokens, true consonant types, NDW	15	Chapman, 2004
Percentage of oral stops*	stable consonant types, percentage of correct consonants		30 (40)	Hardin-Jones & Chapman, 2008
Dental stop tokens		PCC-A	9	Klintö et al., 2014
Observed productive vocabulary (NDW)		NDW	15	Chapman, 2004

*Correlation found in a mixed group of children with and without CP.

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In the screening procedure described by Jørgensen and Willadsen (2017), vocabulary was measured by the Danish version of the CDI (Andersen, Vach, Wehberg, & Bleses, 2006). Consonant inventory measures were assessed by a real-time listening (RTL) procedure (Ramsdell et al., 2012). In RTL, a rater watches an entire video recording in real time without pausing and without taking notes. After watching an entire video recording of a child, the rater writes down consonants and syllable shapes within the child's command. The purpose of RTL is to assess the functional phonetic repertoire a child controls rather than assessing exact phonetic inventory because phonetic transcription greatly overestimates the size of a child's consonant repertoire compared to parent report (Ramsdell et al., 2012). Jørgensen and Willadsen (2017) reported that consonant inventories could be reliably assessed by trained listeners with the RTL procedure in 22 toddlers with CP. Consonant inventory from RTL assessment determined whether predictive consonant measures (oral and true consonants, and oral stops and alveolar stops) were absent or present in participants' consonant inventories. From the CDI it was determined whether participants had small productive vocabularies, defined as a score at or below the 10th percentile. Absence of at least one of the predictive consonant measures and/or the vocabulary measure determined if participants fell into the "need for intervention" category. This evaluation was in almost perfect agreement with the clinical opinion of experienced SLPs, indicating high external validity of the screening procedure. However, as all toddlers produced oral and true consonants, the relevant predictors of need for intervention were a CDI percentile at or below the 10th percentile and absence of alveolar stops which also includes absence of oral stops.

Parents as interventionists

There are many reasons why involving parents in intervention is beneficial. Children spend more time at home than with an interventionist; thus, need and motivation for communication is greater at home, giving rise to more productive learning situations. Perhaps most importantly to families, parent involvement might lead to improved parent self-efficacy in helping their child communicate, thereby reducing anxiety and frustration (Fey, 1986). This has been found in other areas of early intervention other than language (Trivette, Dunst, & Hamby, 2010). Measures of parents' perspectives on parent-implemented interventions have not been reported in earlier studies of toddlers with CP. Studies show that parents can be trained to use language support strategies (e.g. Girolametto, Pearce, & Weitzman, 1996; Roberts & Kaiser, 2015; Scherer et al., 2008), and that parent-implemented intervention affects child language measures positively (e.g. Buschmann et al., 2008; Gibbard et al., 2004; Girolametto & Pearce, 1996). In addition, intervention effects are expected to maintain over time when parents are taught to use language support strategies in naturally occurring interactions with their child, as shown in the studies by Buschmann et al. (2008) and Eiserman, Weber, and McCoun (1992). Two meta-analyses (Law, Garrett, & Nye, 2004; Roberts & Kaiser, 2011) and a recent systematic review (Tosh, Arnott, & Scarinci, 2017) show equal effect of parent-implemented and clinician-implemented early interventions.

Parents can also be involved in evaluation of child speech-language outcomes. Studies have found that parents can make reliable and valid evaluations of reported productive vocabulary (e.g. Dale, 1991; Fenson et al., 1994) and CV syllables (Oller, Eilers, & Basinger, 2001) in children without CP. This is particularly interesting in studies involving babies or toddlers who cannot cooperate with standardised testing. In contrast, parents may report estimates of their child's speech and language use in naturally occurring situations and over time.

Parent-implemented interventions have employed both EMT and FS procedures. Both models use environmental arrangement, responsive interaction, and prompting to facilitate language use (Scherer & Kaiser, 2007). FS uses a high density of modelling while EMT uses modelling as one of several prompting strategies. The most salient difference between the models is that the child is never required to imitate the target form in FS whereas imitation is required in EMT. No studies have compared parent-implemented EMT and FS. Further to this point, Roberts, Kaiser, Wolfe, Bryant, & Spidalieri (2014) compared different language support strategies and found that EMT prompts were hardest for parents to generalise and maintain. Hence, it is likely that FS will be easier for parents to learn and therefore, time for parent instruction can be reduced.

Research questions

The present study aimed to address the gaps in the earlier studies of early intervention in toddlers with CP targeting both vocabulary and consonant inventory directly. Hence, this study investigated if parent-implemented FS intervention targeting vocabulary and consonant inventory directly had an effect on vocabulary and consonant inventory in toddlers with CP and delayed speech and/or language development compared to a matched control group of toddlers with cleft palate who did not receive direct speech-language intervention. Longitudinal results will be collected and reported in a subsequent study.

The study addresses the following experimental questions:

1. Does FS affect gain in productive vocabulary from pre-test to post-test?

2. Does FS affect gain in consonant inventory from pre-test to post-test?

3. Does FS affect need for further intervention?

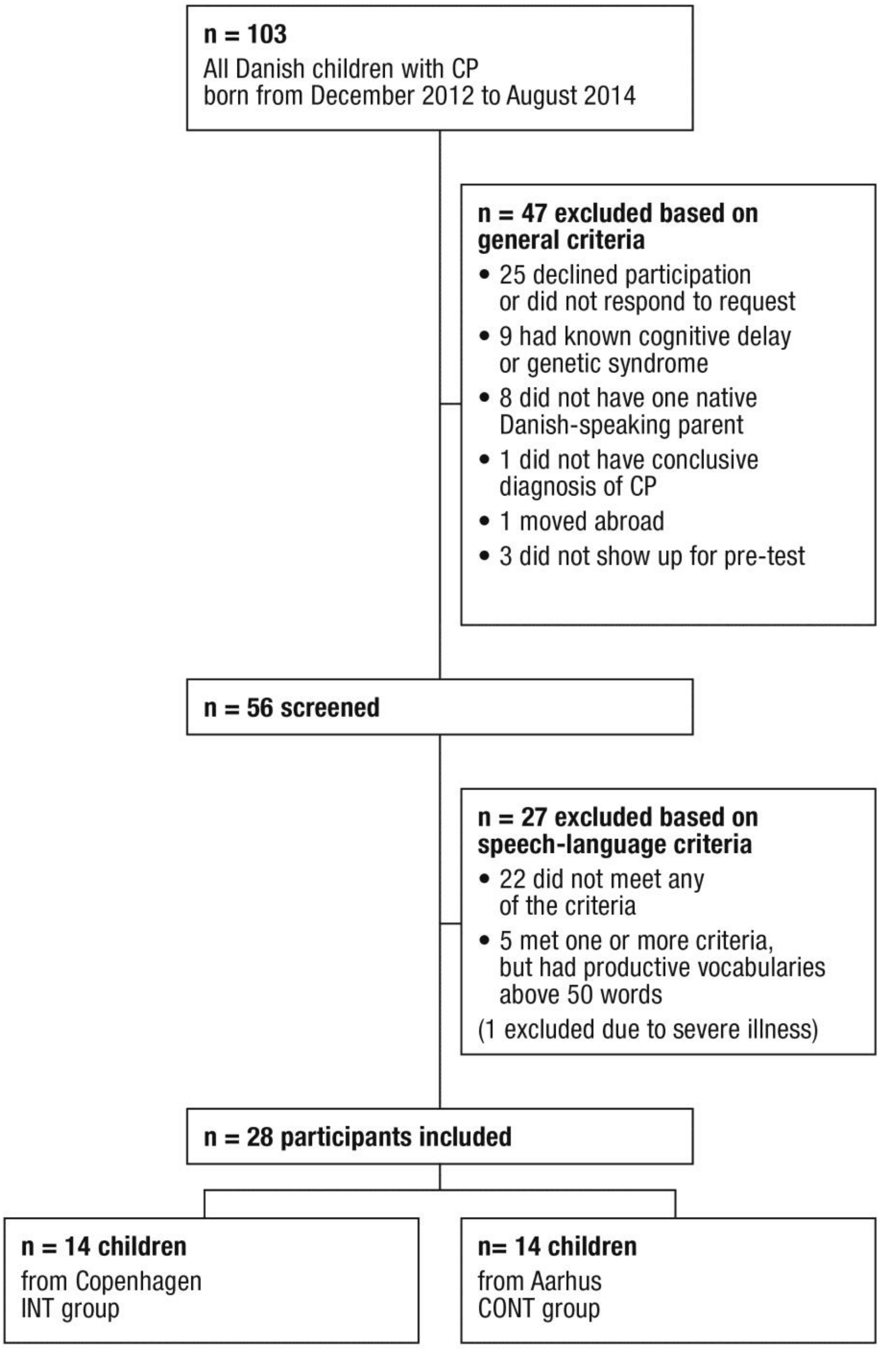
4. What are parents' perspectives on the effects of FS?

Method

Participants

Twenty-eight toddlers with CP, aged 16-24 months, were included in this study. Participants were recruited from a complete birth cohort of all 103 Danish children with CP born between December 2012 and August 2014 via the two cleft palate centres in Denmark. General inclusion criteria were cleft palate with or without cleft lip, at least one native Danish-speaking parent, and absence of a known genetic syndrome, severe cognitive delay, or sensorineural hearing loss. Information was based on parental report and review of medical records. Speech-language inclusion criteria are described below. Fourteen toddlers from Copenhagen cleft palate centre received intervention (INT group), and were matched with fourteen toddlers from Aarhus cleft palate centre who did not receive direct speech-language intervention (CONT group). Eligibility procedures are shown in figure 1.

Figure 1. Flowchart of eligibility procedures.



There were no statistically significant differences between groups for any of the primary matching variables (reported productive vocabulary size, reported productive vocabulary size for age, and age). CDI questionnaires were missing for two participants in the CONT group, but were available from post-test. Therefore, statistical analyses were run again with these participants' scores set at both zero and post-test scores which still revealed no statistically significant group differences. For the secondary matching variables, no significant differences were found for age at palatal surgery, cleft type, or socio-economic status[6] (SES). However, time from palatal surgery to pre-test differed significantly between groups. The INT group had on average had a complete palate for two months longer than the CONT group (see table 2). It was attempted to match participants in pairs on the primary variables. Paired t-tests with BCa bootstrapping revealed good matches for reported productive vocabulary size and reported productive vocabulary size for age, but a significant age difference in the matched pairs. Thus, the matched pairs were kept for the purpose of qualitative evaluation of results, but independent group tests were used for statistical analyses of results.

Table 2. Comparison of pre-test matching variables between the INT group and the CONT group.

	INT group			CONT group			
Primary variables	M (median)	SD	Min-max	M (median)	SD	Min-max	*p*-value
CDI productive vocabulary	14.07 (7.5)	12.45	0-40	10.33 (10.5)	5.85	0-21	.329
CDI percentile	7.86 (5)	8.20	0-25	6.88 (13.75)	8.86	0-30	.772
Age at pre-test	19.36 (19)	2.31	17-24	18.21 (18)	1.42	16-21	.127
Secondary variables							
Age at palatal surgery	13 (14)	2.29	6-16	14 (14)	1.28	12-16	.192
Time from surgery to pre-test	6.36 (6)	2.56	4-12	4 (4)	1.48	2-7	.010*
Cleft type							.436
SES	2.93 (3)	0.91	2-4	3.64 (4)	0.93	2-5	.057

*p<.05

The Bayley III screening subtests of cognitive skills and receptive language (Bayley, 2006) were performed at the pre-test sessions. The cognitive skills subtest was not performed with one participant, but no suspicion of cognitive delay was raised. All participants scored within the 'emergent' or 'competent' categories. Three participants did not comply with the receptive language subtest, and several participants fell into the 'emerging' category because they failed to comply with some of the test items. Characteristics of the 28 participants are shown in table 3, including history of otitis media with effusion (OME) and history of pressure-equalizing

[6] SES is based on highest level of education and current employment status (Ploug, Andersen, Olsen, & Juul, 2012).

tubes (PETs) before 12 months and between 12-18 months. Results from hearing tests were not available.

Written consent to participation was obtained from all parents, complying with ethical principles of the Declaration of Helsinki. The project is registered with the Danish Data Protection Agency.

Design and procedures

All video recordings were made by two experienced SLPs (one in Copenhagen and one in Aarhus). Figure 2 illustrates the study design and procedures.

Pre-tests. All toddlers were seen at two separate pre-test sessions in the clinic. Prior to the first pre-test, parents completed the CDI – Words and gestures. At each pre-test session, natural play interaction between the toddler and one parent was video recorded for 45 minutes. Available toys represented words or could elicit function words used most frequently by Danish toddlers (Wehberg et al., 2007) and represented all Danish consonant phonemes. A list of test items can be seen in appendix 1 and an overview of the Danish consonant system is found in appendix 2. At the second pre-test, the Bayley III screening subtests were performed.

Figure 2. Study design and procedures.

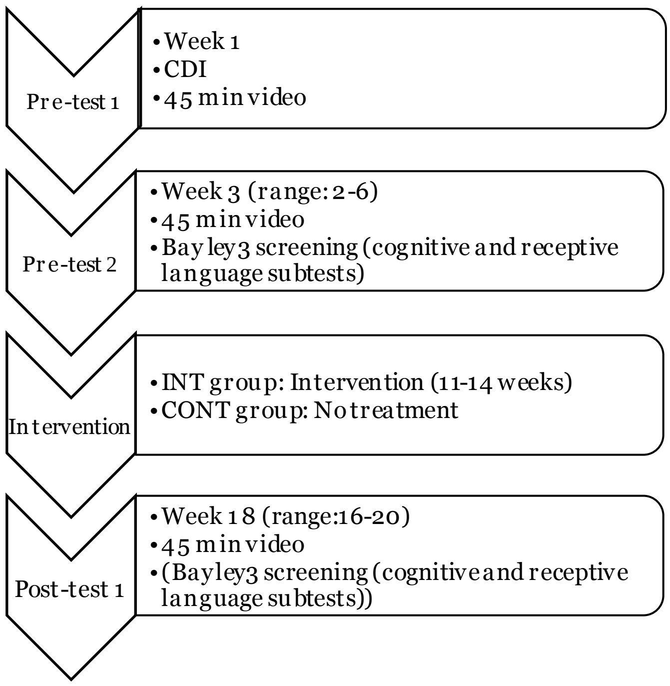


Table 3. Characteristics of the matched participants.

	INT group												CONT group											
Pair	CDI-S	CDI-P	Age at pre-test	SES	Gender	Cleft type	Age at palatal surgery	Time from surgery to pre-test	Bayley3 Cognitive screening	Bayley3 Receptive language screening	History of OME	PETs before 12 m/ PETs 12-18 m	CDI-S	CDI-P	Age at pre-test	SES	Gender	Cleft type	Age at palatal surgery	Time from surgery to pre-test	Bayley3 Cognitive screening	Bayley3 Receptive language screening	History of OME	PETs before 12 m/ PETs 12-18 m
1	17	0	24	4	M	UCLP	14	10	emrg	emrg	y	n/n	10	2.5	20	3	M	BCLP	13	7	emrg	non-comp	y	uk /y
2	25	5	24	2	M	UCLP	14	10	emrg	emrg	y	y/y*	11	2.5	21	3	F	CL/SMCP	-	-	-	cmpt	y	n/n
3	40	25	20	4	M	UCLP	14	6	cmpt	cmpt	y	n/y	8	20	17	3	M	UCLP	12	5	cmpt	cmpt	uk	n/n
4	22	10	20	3	M	CPO	14	6	cmpt	emrg	y	n/y	13	5	19	2	M	CPO	16	3	emrg	emrg	y	y/n
5	9	5	18	3	M	BCLP	12	6	emrg	emrg	y	y/n	18	5	19	4	F	UCLP	14	5	cmpt	emrg	y	n/y
6	5	2.5	17	2	F	BCLP	12	5	cmpt	cmpt	y	y/y	9	2.5	18	5	F	CPO	16	2	emrg	emrg	y	n/y
7	34	25	20	4	M	UCLP	14	6	cmpt	emrg	y	y/y	21	30	18	4	F	CPO	15	3	cmpt	cmpt	y	n/y
8	6	0	20	4	M	UCLP	13	7	emrg	emrg	y	y/y	0	0	18	4	M	UCLP	14	4	emrg	emrg	y	y/y
9	4	5	18	2	M	UCLP	14	4	cmpt	cmpt	y	y/y	11	2.5	19	5	F	CPO	14	5	cmpt	non-comp	y	y/n
10	0	0	18	2	M	CPO	6	12	emrg	emrg	y	n/n**	-	-	17	4	M	UCLP	14	3	cmpt	emrg	y	y/y
11	20	10	20	4	M	UCLP	16	4	cmpt	emrg	y	n/y	14	5	19	2	M	UCLP	14	5	emrg	cmpt	y	y/y
12	5	2.5	18	2	M	UCLP	14	4	cmpt	emrg	y	n/y	5	2.5	18	4	M	CL/SMCP	-	-	cmpt	emrg	n	n/n
13	5	10	17	2	M	UCLP	12	5	cmpt	cmpt	y	n/y	4	5	16	4	M	UCLP	14	2	cmpt	non-comp	y	n/y
14	5	10	17	3	M	CPO	13	4	cmpt	cmpt	y	y/y	-	-	16	4	M	UCLP	12	4	cmpt	cmpt	y	y/n
Means	14.07	8.75	19.36	2.93			13	6.36			14		10.33	10	18.21	3.64			14	4			12	

*PET in one ear. **OME treated with hearing aids from 5-25 months. CDI-S=CDI reported productive vocabulary score, CDI-P=CDI percentile; M=male, F=female; UCLP=unilateral cleft lip and palate, CPO=cleft palate only, BCLP=bilateral cleft lip and palate, CL/SMCP=cleft lip and submucous cleft palate; cmpt=competent, emrg=emerging, non-comp=non-compliant; y=yes, n=no, uk=unknown.

Speech-language inclusion criteria. Participants were included if they matched one or both of the speech-language inclusion criteria: reported productive vocabulary at or below the 10th percentile on the CDI and/or no alveolar stops. These criteria were derived from longitudinal studies of children with CP that found predictive measures of later speech-language performance in toddlers with CP aged 17-21 months (Klintö et al., 2014; Chapman, 2004; Hardin-Jones & Chapman, 2008). In addition, participant must produce less than 50 productive words on the CDI. This criterion was added because intervention aimed at simultaneous expansion of vocabulary and consonant inventory may be most effective in the acquisition of the first 50 words where the interdependency between vocabulary and consonant inventory is strongest (Stoel-Gammon, 2011). The speech-language inclusion criteria were evaluated after the second pre-test. CDI percentile of reported productive vocabulary was determined from the CDI questionnaire, and absence of alveolar stops was assessed by RTL (Jørgensen & Willadsen, 2017; Ramsdell et al., 2012). The 45 minute recordings from pre-test were split into two video files of approximately 22 minutes to account for listener memory constraints, and the rater watched a 22 minute video recording in real time without pausing and without taking notes. Then the rater wrote down CV-syllables within the child's command. The participants produced different amounts of syllables. Keep in mind that unpaused video recordings of 22 minutes were used for RTL assessment. Therefore, a maximum of 662 syllables (produced by the most voluble child in one 22 minutes) and a minimum of 206 syllables (produced by the least voluble child in 90 minutes) per participant were evaluated (see table 4 for an overview of evaluation procedures and table 5 for reliability procedures).

Table 4. Evaluation of consonant inventories at pre-tests and post-test.

Pre-tests	Evaluated units	No. of units Min.-max. (mean, median)	Duration: hh:mm:ss Min.-max. (mean, median)
RTL (56 children)	Min. one video file from pre-tests, max. 662 CV-syllables, as close to 662 CV-syllables as possible	206-662 CV-syllables (459, 460)	00:21:49-01:33:26 (00:59:58, 00:56:55)
Transcription (28 children)	Min. one video file from pre-tests, min. 202 vocalisations	202-504 vocalisations (244, 234)	00:20:24-01:33:26 (00:48:19, 00:44:54)
Post-test			
Transcription (28 children)	Min. one video file from post-test, min. 140 vocalisations	140-455 vocalisations (245, 239)	00:21:07-00:48:45 (00:30:28, 00:23:09)

Post-test. Four months after the first pre-test, parents completed the CDI – Words and sentences before toddlers were video recorded for 45 minutes during natural play with one parent. The same toys/objects as used at the pre-test sessions were available. If a participant failed to comply with the Bayley 3 screening test at pre-test, the test was performed again at post-test.

Transcription

Video recordings were segmented into vocalisations by breath or pause. A vocalisation was defined as minimum one syllabic element, excluding vegetative sounds and fixed signals such as laughter or cry (Oller, Eilers, Neal, & Schwartz, 1999). Vocalisations were transcribed in broad IPA by an SLP with 10 years of experience in transcribing CP speech who was blinded to group assignment. The least voluble participants produced 202 vocalisations at pre-test and 140 at post-test. Hence, these were the minimum numbers of transcribed utterances. Still, to ensure both volume and variability, at least one 22 minute recording was described per participant (see table 4). Table 5 illustrates inter and intra-rater reliability measures for transcription.

Evaluation of word candidates

All vocalisations produced after the first two minutes and within the next 20 minutes of the participants' first pre-test session and post-test were used for evaluation of word candidates. If participants had <50 words on the CDI, word candidates were evaluated using the method proposed by Vihman and McCune (1994). This method is a maximally inclusive identification procedure with a formal system for evaluating word candidates based on both phonetic and contextual criteria. It is however not suitable for children using multiple-word utterances and therefore, participants who produced >50 words on the CDI at post-test were evaluated based on contextual support, parental identification, imitation, and phonetic characteristics as described by Chapman et al. (2003).

Reliability

An overview of inter and intra-rater reliability for all measures is presented in table 5. All measures were re-evaluated by SLPs with 10+ years of experience transcribing the speech of children with CP and/or by trained master's (MA) students. All raters except the first author were blinded to group allocation and time point. The weighted reliability measure (WRM) (Oller & Ramsdell, 2006) was used for evaluation of transcription reliability. Both inter and intra-rater transcription measures were comparable to earlier reports (Oller & Ramsdell, 2006; Preston, Ramsdell, Oller, Edwards, & Tobin, 2011). For intra-rater reliability of observed vocabulary, a wide confidence interval was found for the MA student, indicating variability within evaluations. Nevertheless, the evaluations of the MA student were kept in the analyses since both inter-rater reliability and intra-rater reliability for observed vocabulary were excellent for the main rater.

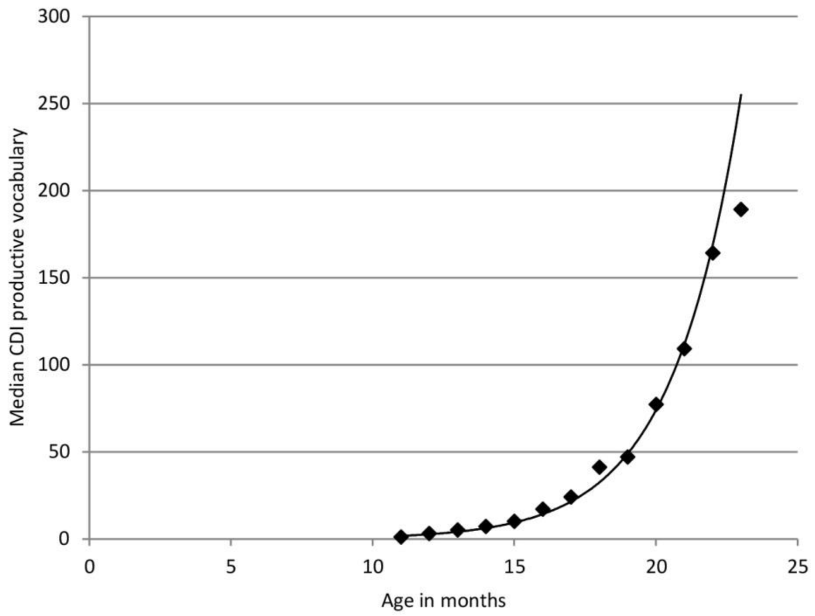
Table 5. Inter and intra-rater reliability across measures.

	RTL	Observed vocabulary	Consonant inventory
Main rater	The first author	The first author 2/3, MA student 1/3	SLP
Secondary rater(s)	SLP, MA student	The first author, MA student	The first author
Re-evaluated material	40% for inter (22 children) 10% for intra (22 children)	25% (across groups and time points)	25% (all children across time points)
Main rater intra	Almost perfect: κ = 0.886	Excellent: ICC = .984, 95% CI [.925-.996]	WRM = 0.88
Secondary rater intra	Substantial to almost perfect: κ = 0.801 and 0.795	Poor to excellent: ICC = .626, 95% CI [-.140-.950]	WRM = 0.88
Inter	Good to excellent: ICC = .743, p<.001, 95% BCa CI [0.70-0.78]	Excellent: ICC = .962, p<.001, 95% CI [.871-.989]	WRM = 0.81

Outcome measures

Productive vocabulary. Gains from pre-test to post-test (reported productive vocabulary) from the CDI as well as number of different words (NDW) participants used in the samples from pre-tests and post-test (observed productive vocabulary). As early productive vocabulary grows exponentially with age, a measure of lexical age was constructed to capture this growth pattern and its possible influence on gain scores. Median productive vocabulary scores from the Danish CDI norms (based on data from 3714 children; Andersen et al., 2006) were plotted against the corresponding age in months, and a growth curve was fitted (see figure 3). From the resulting equation, a CDI-score could be converted into a lexical age score. To convert NDW into a lexical age score, each participant's NDW was multiplied by two to reflect that reported vocabulary is likely to be larger than observed vocabulary (Fenson et al., 1994). Examples of calculations of the lexical age measure are provided in appendix 3. Outcome measures were gain in lexical age for both CDI scores and NDW.

Figure 3. Productive vocabulary growth curve.



Consonant inventory.

Consonant types and tokens in words. Consonant *types* in words included the number of different consonants occurring at least twice in different words (Stoel-Gammon, 1985) whereas consonant *tokens* in words included the total number of consonants in words. If a participant produced more exemplars of a word, the exemplar most phonetically similar to the adult target was evaluated. Only word-initial (WI) consonants were counted. Analysis of consonant inventory based on a minimum number of occurrences is highly sensitive to sample size (e.g. van Severen, van den Berg, Molemans, & Gillis, 2012). However, an increase in use of different consonants as a function of increased observed vocabulary reflects the intervention goal of concurrent expansion of vocabulary and consonant inventory. Accordingly, analyses of consonants in words are based on the criterion of min. two occurrences. A total measure of WI consonant productions was included, but measures of true, oral, oral stop, and alveolar categories included only permissible word-initial (PWI) Danish consonants (see appendix 2).

Consonant types and tokens in all vocalisations. Most participants produced few words at pre-tests; hence, a measure of all vocalisations was included to account for consonant inventory in both prelinguistic vocalisations and words. It was attempted to equalise sample sizes while still retaining the volume and variability of larger samples by bootstrapping the transcribed consonant samples. Bootstrapping is a technique where random re-samples are drawn from an original sample (Efron, 1979), see figure 4. These random re-samples compare to the original sample the way the original sample compares to a population. Bootstrapping has been used to assess consonant inventories in typically developing toddlers (Van Severen et al., 2012) as well as in other areas of child language development (e.g. McKee, Malvern, & Richards, 2000; Xanthos & Gillis, 2010). From each toddler's transcribed consonant sample, a computer script randomly drew 1000 re-samples. The size of the re-sample was determined by the total number of consonants produced by the least voluble participant (n=108 at pre-test; n=139 at post-test). Consonant *types* (total, PWI all, PWI true, PWI oral, PWI oral stops, and PWI alveolar stops) included consonants appearing in at least 95% of 1000 bootstrap samples. Consonant *tokens* (PWI all, PWI true, PWI oral, PWI oral stops, and PWI alveolar stops) included total number of PWI consonants tokens divided by the total number of consonant tokens.

Figure 4. The bootstrapping procedure.

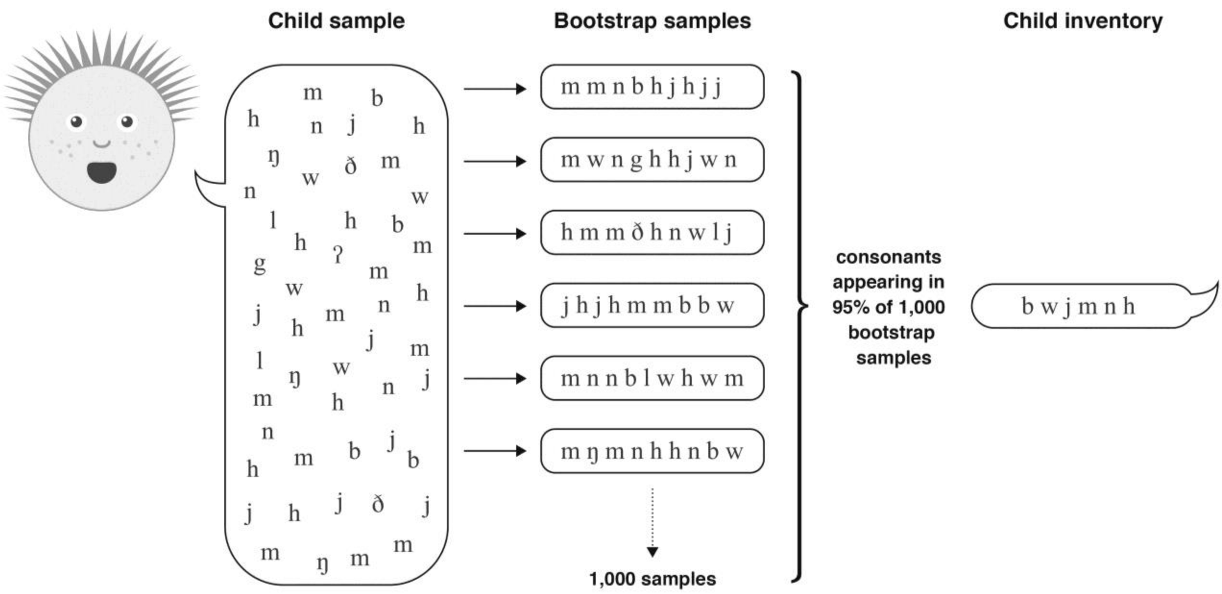


Illustration by Jakob Rønlov.

Need for further intervention. This measure compared how many participants met the speech-language inclusion criteria at post-test in each group. The language inclusion criterion (at or below the 10th percentile for reported productive vocabulary) was assessed from CDI questionnaires at post-test. As participants could score well above the 10th percentile and still produce less than 50 words, the <50 words criterion was not considered at post-test as this criterion only served to ensure that all participants were in the early stage of vocabulary development at pre-test. Unlike the other criteria, the <50 words criterion was not predictive of later speech-language difficulties. The speech inclusion criterion (no alveolar stops) was derived from bootstrapped consonant inventories and consonant inventory in words. The outcome measure was between-group difference in need for further intervention at post-test.

Parents' perspective. At the end of the intervention program, the parents in the INT group were asked to fill out a questionnaire concerning their opinion on their child's progress and their satisfaction with the program. Questions were rated on a four point scale ranging from 1-4 (1: not at all; 2: a little; 3: some; 4: a lot). Outcome measures were parents' assessment of: effect on child measures (vocabulary, speech sounds, sentence lenght, total amount of speech output, interaction), which language support strategies were most effective in changing child behaviour (responsive inteaction, modelling, recasting, phonological emphasis (PE)), and satisfaction with the intervention program concerning use of time, the home-setting, and in general.

The intervention program

The first author carried out all the intervention sessions. Families were seen once a week for six sessions and once every other week for three sessions for a total of nine sessions. Sessions lasted 45-60 minutes, and the total intervention spanned 12 weeks. Parents were encouraged to practice the language support strategies with their child for five minutes every day. Parents in this study were trained to use an FS approach designed to incorporate the recommendations from the meta-analysis by Roberts and Kaiser (2011): intervention sessions were carried out in the families' homes, and parents were taught responsive interaction, modelling of target words, and expansions, including speech recasts in which phonological information was added in immediate response to a child's words (Scherer & Kaiser, 2010). A description of the intervention program is shown in appendix 4. Parents received written information on the rationale and over-all content of the intervention before the intervention began. Every intervention session contained all elements of the teach-model-coach-review instructional approach (Roberts et al., 2014). Each session began with reviewing last session's homework plan. Then, parents were informed about a language support strategy and provided with an explanation of the strategy's implication for their child's speech and language development (the teach component, 10-15 minutes). Afterwards, the first author demonstrated use of the strategy with the child (the model component, 5-10 minutes). The parents practised using the language support strategy with their child while the first author video recorded the interaction for 5-10 minutes. Coaching was provided during the interaction if parents did not use the strategy or seemed uncomfortable in the interaction; otherwise, coaching was provided immediately after interaction via the video recording (the coach component, 10-15 minutes). The indirect coaching provided an opportunity to watch parts of the interaction more than once and enabled parents to reflect on their use of strategies. Also, it reduced the amount of time the child had to cooperate. Finally, homework was planned together with parents (the review component, 5-10 minutes). Parents wrote down how and when they would use a strategy. They were required to keep a log of when and for how long they practiced, and how their child responded, including production of target words. Furthermore, parents were encouraged to send audio recordings of one practice session a week to the author. Both logs and recordings were used to evaluate homework in the following intervention session.

Intervention targets. Target words were selected in cooperation with parents based on criteria described by Lahey and Bloom (1977) and Wilcox, Kouri, and Caswell (1991): 1. Words that the child comprehended but not yet produced based on information from the CDI. 2. Words beginning with sounds within the child's consonant inventory. This information was derived from the RTL procedure. 3. Words that could be demonstrated by objects or functions during play or everyday activities. 4. Words that occur frequently in early vocabularies. Consonant inventory targets were incorporated by adding words beginning with sounds outside the child's consonant inventory. The child's interests and home environment was considered in selecting word targets. None of the toys/objects available at pre and post-test were used as intervention targets. A maximum of ten target words were trained at a time. When a participant produced a target word at least twice in an intervention session, or when parents reported that their child produced a target word in-between sessions, a new target word was introduced.

Treatment fidelity

Parental attendance was registered, and two families attended seven and eight home visits respectively, while 12 families attended all nine home visits. Due to families' situations, the total duration of the intervention program ranged from 11-14 weeks.

Parent behaviours were examined during the intervention to ensure learning of specific strategies and to set goals for the next intervention session. Responsiveness, modelling, and expansions including speech recasts were evaluated in the intervention session following introduction. Responsiveness was defined as a contingent parent response within two seconds of the child's vocalisation. Expansions including speech recasts were broadly defined as repeating some or all of a child's words with addition of syntactic, semantic and/or phonological information (Cleave, Becker, Curran, Van Horne, & Fey, 2015). Parent-child interaction in five minutes was video recorded, and parents had to demonstrate 75% responsiveness, use of expansions in 40% of responses, and more than two models of at least three target words. Training on one strategy continued until criterion levels were met. A second SLP evaluated criterion levels at the end of the intervention program from intervention video recordings, showing that all parents met the criterion levels for responsiveness and modelling. Eleven of 14 parents met the criterion for expansions (mean: 55%, min-max: 27-93%).

Parent self-evaluation was explored qualitatively from parents' weekly log of their homework and from a parent questionnaire answered at the end of the intervention program. Logs revealed that six parents practised at least five times per week during the first four weeks, six parents practised at least three times, and two parents practised 0-2 times per week. After the first month of the program, all parents increased their use of strategies in the home, especially in everyday activities. In the questionnaire, nine of 14 parents reported that they did homework five times a week, and 12/14 parents reported doing homework for at least 30 minutes per week in total. All but one parent reported practise of language support strategies a lot or to some extent in both the planned homework activities and in everyday situations. Several parents commented that they practised increasingly during the intervention as they became more familiar with the strategies.

The CONT group

In Denmark, standard speech-language treatment for babies and toddlers with CP consists of parent counselling in the cleft clinic, usually at ages ten, 18, 24, and 36 months. The specialist SLP may refer the family to local speech-language therapy services; however, early speech-language intervention is uncommon in Denmark. In this study, the CONT group did not receive any speech-language intervention.

Analyses

Statistical analyses were performed using SPSS statistical package version 22 (SPSS Inc., Chicago, IL). In many cases, the data violated assumptions of normality. Therefore robust independent samples t-tests with bias-corrected and accelerated bootstrapping (BCa) were used in between-group analyses while robust paired samples t-tests with BCa bootstrapping were used in pre-post analyses. Mann Whitney U tests or Wilcoxon signed rank tests were used in cases where BCa bootstrapping could not be performed. To account for pre-test differences, gain scores were used. Negative gain scores were counted as zero change. ICC estimates and their 95% confidence intervals were calculated based on a mean-rating, absolute-agreement, two-way random-effects model. Single measures are reported for both inter-rater and intra-rater reliability. The magnitude of difference between independent variables cannot be determined by p-values alone (see Durlak, 2009). Furthermore, p-values are reflective of sample size. Therefore, effect sizes are reported along with p-values in this study. Interpretations of effect sizes and reliability measures are based on commonly cited benchmarks (Cohen's *d*: .2 small, .5 medium, .8 large (Cohen, 1988); Kappa: < 0 no agreement, 0-.20 slight, .21-.40 fair, .41-.60 medium, .61-.80 substantial, and .81-1 almost perfect (Landis & Koch, 1977); ICC: <.40 poor, .40-.59 fair, .60-.74 good, and .75-1.0 excellent (Cicchetti, 1994) as well as clinical relevance.

Results

Pre-test scores in the compared groups

No statistically significant differences were found between groups for any of the outcome measures at pre-test (see table 6).

Table 6. Between-group comparisons of outcome measures at pre-test.

Outcome measure	INT group			CONT group			*p*
	M (median)	SD	min-max	M (median)	SD	min-max	
Productive vocabulary							
CDI score+	14.07 (7.5)	12.45	0-40	9.64 (10)	5.59	0-21	.249
Lexical age CDI score+	15.01 (14.44)	2.41	10-18.47	14.61 (15.17)	1.88	10-16.94	.628
NDW	5.86 (4)	4.26	0-13	6.29 (5)	2.95	3-14	.760
Lexical age NDW	14.85 (14.64)	2.12	10.00-17.45	15.51 (15.17))	0.99	13.96-17.62	.296
Consonant inventory in words							
All types	0.71 (0)	0.91	0-2	0.86 (1)	0.86	0-3	.674
PWI types	0.71 (0)	0.91	0-2	0.86 (1)	0.86	0-3	.674
PWI true types	0.43 (0)	0.65	0-2	0.43 (0)	0.51	0-1	1.000
PWI oral types	0.21 (0)	0.43	0-1	0.29 (0)	0.47	0-1	.676
PWI oral stop types	0.21 (0)	0.43	0-1	0.64 (0)	0.36	0-1	.637
PWI alveolar stop types	0.00			0.07 (0)	0.27	0-1	.125
PWI tokens	1.64 (0)	2.13	0-6	1.86 (2)	1.79	0-4	.776
PWI true tokens	1.00 (0)	1.71	0-6	1.00 (0)	1.24	0-3	1.000
PWI oral tokens	0.50 (0)	1.02	0-3	0.64 (0)	1.08	0-3	1.000
PWI oral stop tokens	0.50 (0)	1.02	0-3	0.36 (0)	0.93	0-3	.701
PWI alv stop tokens	0.00			0.21 (0)	0.80	0-3	.654*
Consonant inventory in all vocalisations							
All types	6.79 (7)	1.31	5-10	6.57 (7)	1.70	4-9	.712
PWI types	5.93 (6)	1.07	4-8	5.79 (5.5)	1.25	4-8	.748
PWI true types	3.07 (3)	1.00	1-5	2.93 (3)	1.00	1-4	.708
PWI oral types	3.50 (3.5)	1.22	1-6	3.29 (3)	1.20	2-6	.645
PWI oral stop types	1.07 (1)	0.92	0-2	0.93 (1)	0.83	0-2	.669
PWI alveolar stop types	0.14 (0)	0.36	0-1	0.29 (0)	0.47	0-1	.376
PWI tokens	86.72 (88.00)	7.76	67.15-94.59	86.97 (87.20)	5.65	75.52-94.87	.930
PWI true tokens	36.74 (40.46)	13.87	15.58-55.00	38.56 (43.59)	13.95	11.11-57.94	.708
PWI oral tokens	43.39 (44.78)	15.19	13.57-63.08	35.17 (34.46)	17.54	8.29-67.70	.223
PWI oral stop tokens	11.55 (6.89)	14.59	0-47.91	7.17 (3.13)	11.21	0-40.45	.383
PWI alv stop tokens	1.43 (0)	4.48	0-16.70	1.46 (0)	2.60	0-7.02	.989

*Kruskall-Wallis test. +Results from two CONT participants missing.

Target words

The mean number of presented words was 22 (min-max: 13-30). Seventy per cent of the target words contained consonants outside pre-test inventory (min-max: 23-85%). Only target words of one participant contained less than 50% outside-inventory consonants because this participant had a large pre-test inventory. The mean number of learned target words was 11 (min-max: 3-21). Fifty-three per cent of the words with outside-inventory consonants and 57% of the words with within-inventory consonants were learned.

Productive vocabulary

CDI scores. Gain scores could not be computed for three CONT participants because CDI questionnaires were missing for one of the time points. Thus, results from these participants as well as their matched counterparts in the INT group are not reported here. Independent t-tests with BCa compared gain in CDI-score and gain in lexical age for CDI scores from pre-test to post-test between groups. As seen in table 7, no statistically significant differences were found.

Table 7. Between-group comparison of gain in productive vocabulary measures from pre-test to post-test.

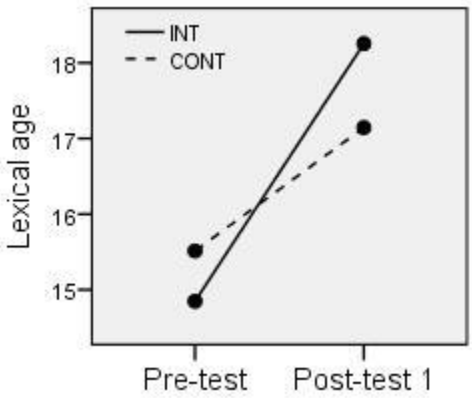
Gain scores	INT group			CONT group			p	d
	M (median)	SD	min-max	M (median)	SD	min-max		
CDI+	57.27 (52)	21.45	16-92	54.72 (30)	67.59	8-245	.918	0.05
Lexical age CDI+	4.21 (3.86)	1.92	0.92-6.69	4.17 (4.32)	2.02	1.47-7.79	.950	0.02
NDW	16.86 (19.5)	9.77	2-30	9.14 (8)	9.72	0-29	.059	0.79
Lexical age NDW	3.41 (2.98)	1.45	1.30-6.53	1.69 (1.91)	1.38	0-3.99	.009	1.21¤

*$p<.01$, ¤ large effect size. +Results from 22 participants.

NDW. Independent t-tests with BCa compared NDW gain and lexical age gain for NDW from pre-test to post-test between groups. The INT group showed a greater NDW gain (mean change: 16.86 words) than the CONT group (mean change: 9.14 words); however, the difference was not statistically significant (see table 7). A statistically significant difference with a large effect size was found for NDW lexical age between the INT group (mean change: 3.41 months) and the CONT group (mean change: 1.69 months): $t(26)=3.201$, $p=0.009$, 95% BCa CI [0.70-2.83] , d=1.21 (see figure 5).

CDI scores, NDW, and their corresponding lexical age scores from all participants are presented in appendix 5.

Figure 5. Between-group difference in NDW lexical age gain.



Consonant inventory

Consonant types and tokens in words. Independent t-tests with BCa compared gain in total and permissible word-initial (PWI) consonant types and tokens in words from pre-test to post-test between groups. Because BCa could not be performed for gain in PWI alveolar stop types in words, a Mann-Whitney U test was performed for this measure. There was a statistically significant difference with a large effect size in gain in PWI true consonant types in words between the INT group (mean change: 2.36 true consonant types) and the CONT group (mean change: 1.21 true consonant types): $t(21.731)=2.147$, $p=0.043$, 95% BCa CI [0.04-2.19], $d=0.81$ (see figure 6).

The INT group also showed greater mean gain scores for all other tested consonant type and token measures than the CONT group (see table 8), but these differences were not statistically significant. However, a large effect size was found for gain in oral stop tokens.

Figure 6. Between-group difference in gain in true consonants in words.

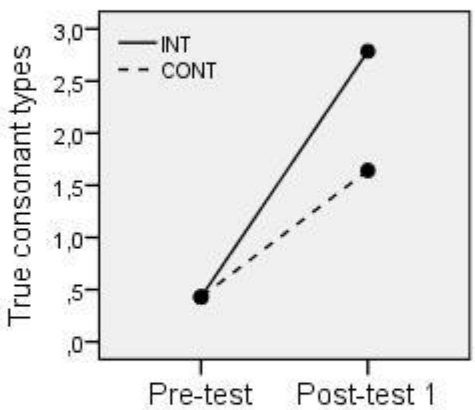


Table 8. Between-group comparison of gain in consonant type and token measures in words from pre-test to post-test.

Consonant type measure	INT group			CONT group			p	d
	M (median)	SD	min-max	M (median)	SD	min-max		
Total	3.00 (3)	2.04	0-6	2.21 (2)	1.63	0-5	.270	0.43
PWI all	2.86 (3)	2.07	0-6	1.93 (1.5)	1.64	0-5	.200	0.50
PWI true	2.36 (2.5)	1.69	0-5	1.21 (1)	1.05	0-3	.043*	0.81¤
PWI oral	1.57 (1)	1.55	0-4	1.21 (1)	1.25	0-4	.509	0.25
PWI oral stops	1.43 (1)	1.34	0-4	0.64 (0)	0.84	0-2	.075	0.70
PWI alveolar stops	0.21 (0)	0.43	0-1	0.14 (0)	0.53	0-2	.345	0.22
Consonant token measure								
PWI all	11.86 (10.5)	9.03	0-25	8.29 (6)	8.53	0-27	.288	0.41
PWI true	9.64 (8.5)	7.64	0-23	5.14 (4.5)	4.94	0-14	.100	0.68
PWI oral	6.43 (4)	7.25	0-19	3.29 (2.5)	3.69	0-10	.167	0.56
PWI oral stops	6.14 (4)	6.76	0-18	1.79 (0)	2.86	0-9	.060	0.86¤
PWI alveolar stops	0.93 (0)	1.69	0-5	0.36 (0)	1.34	0-5	.225	0.47

*p<.05, ¤ large effect size.

Consonant inventories from all participants are presented in appendix 6.

Consonant types and tokens in all vocalisations. Independent t-test with BCa compared gain in consonant types and tokens in all vocalisations from pre-test to post-test between groups. Table 9 shows that the INT group had greater mean gain scores than the CONT group for all measures except true and oral consonant tokens. None of these differences were statistically significant. Effect sizes ranged from non-existent to medium.

Table 9. Between-group comparison of gain in consonant type and token measures in all utterances from pre-test to post-test.

Consonant type measure	INT group			CONT group			*p*	*d*
	M (median)	SD	min-max	M (median)	SD	min-max		
Total	2.79 (3)	2.25	0-6	1.79 (1.5)	1.67	0-5	.182	0.52
PWI all	2.64 (2.5)	1.98	0-6	1.43 (1)	1.40	0-4	.073	0.71
PWI true	2.50 (2)	2.07	0-6	1.36 (1)	1.34	0-4	.094	0.66
PWI oral	2.14 (2)	1.79	0-5	1.29 (1.5)	1.19	0-3	.143	0.57
PWI oral stops	1.71 (2)	1.20	0-3	0.64 (0)	0.84	0-3	.159	0.55
PWI alv stops	0.57 (0.5)	0.65	0-2	0.36 (0)	0.50	0-2	.335	0.37
Consonant token measure								
PWI all	5.90 (2.57)	7.29	0-19.20	3.08 (0.81)	3.69	0-9.54	.241	0.49
PWI true	48.04 (26.42)	56.93	0-182.52	55.08 (19.21)	100.73	0-376.63	.818	-0.09
PWI oral	48.27 (10.88)	92.78	0-341.73	56.07 (13.84)	100.77	0-359.96	.850	-0.08
PWI oral stops	211.80 (122.45)	305.69	0-1167.50	154.73 (71.67)	201.48	0-630.50	.563	0.22
PWI alv stops	125.33 (4.57)	182.63	0-490.00	124.56 (20.76)	204.55	0-736.00	.993	0.00

Need for further intervention

A Fischer's exact test compared need for intervention between groups at post-test. Six out of 14 INT toddlers and 12 out of 14 CONT toddlers needed further intervention. This difference was statistically significant, $\chi^2(1)=5.60$, $p=0.018$. Based on the odds ratio, the INT group was eight times more likely not to need intervention at post-test.

Parents' perspective

All parents reported that the intervention program in general affected their child's speech positively (mean: 3.8; min-max: 3-4). The largest effects were reported for vocabulary and interaction (mean: 3.6; min-max: 3-4 for both measures), followed by speech sounds and total amount of speech output (mean: 3.4; min-max: 2-4 for both measures). The smallest effect was seen for sentence length (mean: 3.0; min-max: 2-4). Recasting was believed to be the most effective language support strategy in changing child behaviour (mean: 3.9; min-max: 3-4) followed by modelling (mean: 3.8; min-max: 3-4) and PE (mean: 3.6; min-max: 3-4). All parents reported that the amount of time they had to spend on testing, the intervention program, and homework was appropriate, and they expressed high satisfaction with the intervention being home-based. In general, parents were highly satisfied with the program (mean: 3.8; min-max: 3-4). Parents made comments that the program made them feel more capable to understand and interact with their child, and this helped their child to communicate better and in turn express less frustration.

Post-hoc tests

The mixed productive vocabulary findings (statistically significant differences for gain in NDW lexical age, but not for CDI measures) lead to a posthoc investigation of correlations between the CDI and NDW measures. Pearson product-moment correlation coefficients revealed a strong significant correlation between CDI scores and NDW at pre-test, $r(20)=.652$, $p<.001$), and also between CDI lexical age and NDW lexical age at pre-test, $r(20)=.556$, $p=.007$). However, no significant correlations were found for these measures at post-test ($r_s(20)=.156$, $p=.489$ for both comparisons). Correlations in each group at post-test were also non-significant (INT: ($r_s(20)=.343$, $p=.304$ for both comparisons; CONT: $r_s(20)=-.123$, $p=.718$ for both comparisons).

The observed intervention effect on gain in true consonant types in words did not seem to be a result of consonant inventory expansion as no significant differences or large effect sizes were found for any between-group measure of consonants in all vocalisations. Therefore, it was investigated whether the observed intervention effect on gain in true consonant types in words was caused by INT toddlers learning to use true consonant types already within their inventory at pre-test (IN-sounds). Follow-up testing showed that in words at post-test, INT toddlers used 63 % of the true consonant types they produced in all vocalisations at pre-test. The CONT group only used 37% (see appendix 6). A multiple linear regression was calculated to predict true consonant gain in words. Regression was based on the proportion of true consonant types used in words at post-test that were also present as true consonant types in all vocalisations at pre-test. Gain from pre to post-test in true consonant types in all vocalisations was added to the model as a second independent variable. A statistically significant regression equation was found ($F(1,26)=42.302$, $p<.001$) with an R^2 of .619. Both independent variables were statistically significant predictors of gain in true consonant types in words, but gain in true consonant types in all vocalisations only added .062 to the explained variance.

Discussion

This study aimed to investigate the effect of focused stimulation on observed vocabulary and consonant inventory in toddlers with CP and delayed speech and/or language development. Results were compared to a matched control group of toddlers with CP who did not receive direct speech-language intervention.

All but two between-group comparisons of both vocabulary and consonant inventory measures revealed higher gain scores in the INT group than in the CONT group. In addition, effect sizes were medium to large in half of the vocabulary and consonant inventory measures. However, only statistically significant differences will be discussed below, unless otherwise stated. The

INT group learned target words with target consonants outside inventory and within inventory at similar rates, suggesting that both vocabulary and consonant inventory are viable intervention targets for toddlers with CP. Increase in most measures was evident whether or not the participants received intervention which underlines the importance of including matched control groups in intervention studies of toddlers with CP.

Productive vocabulary

The INT group obtained significantly larger NDW lexical age gain scores than the CONT group. A large effect size implies a clinically significant magnitude of difference between groups. The clinical significance is further underlined by the result that gain scores were higher in the INT toddler in 11/14 cases. Lexical age gain in months does not directly correspond to typical vocabulary development in months. The lexical age measure was calculated from each participant's NDW but derived from median reported productive vocabulary scores from the Danish CDI norms. To reflect the expected difference between observed and reported vocabulary size, each participant's NDW from the observed vocabulary was multiplied by 2 (Fenson et al., 1994). The mean gain of 3.41 months in the INT group resembles typical productive vocabulary development since post-test was performed four months after pre-test. The mean gain of 1.69 months in the CONT group seems to be in agreement with reported results of earlier studies that found slower vocabulary growth of toddlers with CP compared to their peers without CP (Broen et al., 1998; Lu et al., 2010; Hardin-Jones & Chapman, 2014; Willadsen, 2012). It would be a clinically significant result for toddlers with CP to obtain typical productive vocabulary growth trajectories. Interestingly, the highest gain in a CONT participant was 3.99 which would correspond to normal lexical growth in four months and may thus indicate that no CONT participant expanded NDW beyond general development. Hence, a measure of lexical age seems well-suited to capture NDW change in toddlers with CP and small vocabularies because of exponential vocabulary growth patterns. In contrast, no intervention effects were seen for CDI productive vocabulary scores. Posthoc tests revealed that CDI scores and NDW were associated at pre-test, but not at post-test. This raised the question if parent training improved the validity of parent-reported measures as also suggested by Feldman et al. (2000) and Girolametto et al., (1996). However, non-significant correlations between CDI and NDW were found in both groups at post-test which suggests that absence of an association could not be explained by superior evaluations by INT parents. Nonetheless, other factors may have affected the parent-reported outcomes which raises the question whether the CDI is valid measure of treatment effect. Another explanation for the observed gain in lexical age gain may be that INT parents were trained to facilitate child communication. Thus the observed intervention effect on NDW gain may be related to parents' ability to facilitate child word production at post-test, but not necessarily overall productive vocabulary. Longitudinal results will provide insights into whether parents' ability to facilitate word use will also affect overall vocabulary over time. The lexical age measure is similar to an age-equivalent score. Age-equivalent scores are widely criticised mainly because they fail to account for the range of normal performance, in contrast to standard scores and percentile ranks (Lawrence, 1992). However, the lexical age measure is solely developed to measure change in the early vocabulary and not intended as a norm reference in clinical evaluation.

Consonant inventory

A significant difference with a large effect size was found in gain in PWI true consonant types in words between the INT group and the CONT group. Clinical significance is further emphasized by comparing the gain scores of the 14 matched pairs in which the INT toddler had the higher gain score in nine cases whereas this was only true for two CONT toddlers. Number of true consonant types in words is potentially clinically relevant as Chapman (2004) found that for children with CP, true consonant types in words at 21 months is correlated with NDW at 39 months. In addition, use of true consonants has been linked to later phonological proficiency (Vihman & Greenlee, 1987) and vocabulary size (Stoel-Gammon, 1989) in children without CP. While a difference of one true consonant type between groups (mean change INT: 2.36; CONT: 1.21) may not appear to be of great importance for communication ability, it is important to consider that participants on average did not even produce one true consonant type in words at pre-test (mean: 0.43 in both groups, see table 6). For a toddler, being able to use words beginning with e.g. both /b/ and /m/, compared to just one of these consonants, may significantly impact both vocabulary size and intelligibility. For instance, of the first 50 words acquired in Danish toddlers, nine words begin with /b/ and seven words begin with /m/ (Wehberg et al., 2007). Thus, the ability to use both /b/ and /m/ almost doubles the size of a participant's potential early vocabulary. Post-hoc testing showed that a large part of the variance in gain in true consonant types in words was explained by INT toddlers having learned to use true consonant types in words which were already within their inventory at pre-test. This means that FS may aid toddlers in using their already well-practiced consonants in words. Toddlers with CP and small vocabularies may not be able to use their prelinguistic consonant inventories in words to the same extent as TD toddlers (Vihman et al., 1985). Interestingly, it has been suggested that small vocabularies in toddlers may not only be related to cognitive-linguistic delay or restricted consonant inventories, but also in some toddlers may be caused by inability to use prelinguistic inventory in words (Stoel-Gammon, 1998b; Vihman, 1992). Nonetheless, studies have found patterns of lexical selectivity in the early words of toddlers with CP (Estrem & Broen, 1989; Hardin-Jones & Chapman, 2014; Willadsen, 2013). Thus, the results of this study may indicate that a subgroup of toddlers with CP and speech-language delay have specific difficulties with using their consonant inventories in words.

Another explanation may be that the consonant inventories of toddlers with CP are probably not as well-practiced as consonant inventories in TD toddlers because of the restrictions posed by the open palate before surgery. The 'articulatory filter' hypothesis assumes that children unconsciously filter adult input through well-practised productions (see Vihman, 2017). Thus, consonant inventories in these toddlers with CP may not be practised enough for the 'articulatory filter' to work. Further, length of time over which a toddler has been practicing consonants does seem to correlate with later production ability (Keren-Portnoy, Vihman, DePaolis, Whitaker, & Williams, 2010). The saliency of within-inventory consonants presented frequently by parents in target words may be responsible for establishing a link between consonant inventory and words. If this is the case, then target words should mainly contain within-inventory consonants. Future intervention studies could investigate the effects of targeting consonants within and outside inventory in intervention by manipulating these in target words.

Although not statistically significant, a large effect size (d=0.84) was found between the INT group and the CONT group for oral stop tokens in words. Clinical significance was further emphasized by comparing the gain scores of the 14 matched pairs where the child in the INT group had the higher gain score in eight cases whereas this was only true in three cases for the CONT group. Most children produced very few oral stop tokens in words at pre-test (INT mean: 0.50; CONT mean: 0.36, see table 6). At post-test, the INT group produced 6.64 oral stop tokens in 22.72 words while the CONT group produced 2.15 oral stop tokens in 15.43 words (mean values). Almost half of the words produced by 80% of Danish toddlers at 24 months begin with a stop consonant (Ingólfsdóttir, 2012); thus, being able to produce more stops may impact both vocabulary size and intelligibility. Further, the number of oral stop tokens may hold clinical relevance as one study of toddlers with CP (Hardin-Jones & Chapman, 2008) found a medium sized correlation between oral stop production at 17 months and measures of consonant accuracy at 27 month.

The bootstrapping procedure enabled comparison of consonant inventories in all vocalisations between participants, but it may have under-estimated the consonant inventories in the more voluble individual participants. This is demonstrated by comparing bootstrapped consonant inventories in spontaneous speech to consonant inventories in words. In words, consonants were present if they appeared at least twice in different words. As seen in appendix 6, some participants produced consonants in words that were not present in their bootstrapped inventories in all vocalisations. This was true for five INT participants and two CONT participants and may thus have decreased inventories more in the INT group. However, establishing consonant inventories in both linguistic and prelinguistic vocalisations is a compromise between valid group comparisons and retaining information from individual participants.

A note on velopharyngeal dysfunction (VPD)

VPD is notoriously difficult to detect in toddlers, but absence of oral stops more than two months after palatal surgery in toddlers with CP may indicate VPD, even though absence of oral stops in itself cannot confirm VPD. Four INT toddlers and eight CONT toddlers produced no oral stops at pre-test according to the screening procedure. Interestingly, of these participants, only one INT toddler produced oral stops in words at post-test, whereas the remaining 11 participants still did not produce oral stops in words. It is likely that some of the participants who did not produce oral stops may later be diagnosed with VPD, and that underlying VPD may have influenced the results of this study. In three of the matched pairs (1, 11, and 14; see appendix 6), none of the participants produced oral stops at pre-test. In pair 1, the INT toddler had higher gain scores for vocabulary measures and true consonant types than the CONT toddler. In pair 11, the INT toddler had higher gain scores for true consonant types and oral stop tokens while the CONT toddler had higher scores for vocabulary measures. In pair 14, the INT toddler had higher gain scores for vocabulary measures. In addition, gain scores for INT participant 1 (vocabulary measures) and for INT participant 11 (true consonant types) were high compared to all participants' gain scores for these measures. Although this is not a clear-cut picture, it indicates that intervention can be beneficial to toddlers with CP in the presence of underlying VPD. Long-term results may shed light on whether intervention gain is predictive of a later VPD diagnosis.

Need for further intervention

The INT group were eight times more likely to not need intervention at post-test than the CONT group. This result is perhaps the most compelling result as the speech-language inclusion criteria were derived from measures obtained at 17-21 months correlating with later measures of speech accuracy and vocabulary in 3-year-olds in longitudinal studies of children with CP (Klintö et al., 2014; Chapman, 2004). Thus, not matching the speech-language inclusion criteria may have positive implications for future speech-language development.

Parent outcomes

Parents were able to learn language support strategies; yet, not all parents used expansions including recasts at criterion level in the maintenance phase. This may have been caused by a low number of word productions for parents to expand or recast and/or parents' inability to recognise these productions as words. Speech recasting may be more efficient for children with larger vocabularies whereas children with smaller vocabularies may benefit more from responsive interaction and use of target words, as also pointed out by Kaiser et al. (2017).

In general, parents reported that the intervention program positively affected their child's speech-language development with largest perceived effects reported for vocabulary and interaction, followed by speech sounds and total amount of speech output and the smallest effect for sentence length. Four parents reported little effect on sentence length. Interestingly, three of these toddlers were the only INT participants who did not reach 50 productive words according to parent report at post-test whereas the last participant produced 72 words. This lends credibility to parents' observations of sentence length as two-word utterances typically appear when children have acquired a productive vocabulary of 50 words (Brown, 1973). Similarly, two parents reported little effect on their child's speech sounds. These were the participants with the smallest consonant inventories in all vocalisations at post-test, and they were two out of three participants who did not produce oral stops in words. Thus, parents' perspective seem to mirror the outcome measures of vocabulary and consonant inventory but possibly also reflects which measures are more important to parents. Overall, it seems that parents' perspective is not only valuable as a general measure of the impact of early intervention on child communication ability: parents may also be able to specifically identify speech-language outcome measures. This is not surprising as several studies have found parents to be reliable and valid reporters of the speech-language abilities of infants and toddlers without CP (e.g. Dale, 1991; Fenson et al., 1994; Oller et al., 2001). In line with the recommendations for parent-implemented early intervention (Roberts & Kaiser, 2011), parents found that recasting and modelling were the most efficient strategies for changing child behaviour. Parents were satisfied with the intervention being home-based and the total duration of the intervention program, indicating that the intervention program is feasible in families with two working parents. One parent specifically reported that the family would not have been able to attend sessions had they been carried out in the cleft centre. Furthermore, comments from parents indicate that the intervention program not only affected child behaviour, but also increased parents' self-efficacy in understanding and interacting with their child.

Limitations

Group sizes were relatively small which may explain why few results in the present study were statistically significant, even though medium to large effect sizes were found for most between-group comparisons. Analyses involving CDI measures are limited because they only involved comparison of 11 participants in each group due to missing data. Participant matching was not complete. In particular, it was concerning that the CONT group had had a complete palate for a significantly shorter period of time than the INT group. This may have favoured gains in the INT group. On the other hand, the INT group had had longer time to speak with a complete palate but still evidenced need for intervention at pre-test. Thus, the INT group may actually have had a greater need for intervention than the CONT group. However, all participants had had at least two months to adjust to speaking with a complete palate as suggested by Grunwell and Russell (1987). Participants with mixed cleft types were included to enrol as many participants as possible. In general, studies have reported that speech is more affected in more extensive clefts (Hardin-Jones, Chapman, & Schulte, 2003; Lohmander-Agerskov, Söderpalm, Friede, Persson, & Lilja, 1994; Persson, Elander, Lohmander-Agerskov, & Söderpalm, 2002). The issues of mixed cleft types and heterogeneity were to some extent addressed by introducing speech-language criteria.

Receptive language is difficult to test reliably in toddlers, and Danish tests for children below two years of age do not exist. Therefore, the screening version of Bayley3 was used. As it is a screening tool, the receptive language levels of some of the participants may have been over- or underestimated. In addition, two toddlers in the CONT group declined participation in the receptive language subtest which opens the possibility that these participants did not meet the general inclusion criteria. Hearing tests were not performed which may also have influenced results. However, presence of OME and early PET insertion were highly similar in the two groups.

Conclusion

Parent-implemented focused stimulation positively affected lexical age, a measure of observed vocabulary, true consonant inventory in words, and need for intervention in this study of toddlers with CP and delayed speech and/or language development. Parents were generally satisfied with the intervention program. Longitudinal results will be obtained and reported in a separate study.

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Appendix 1. Available toys or function words at pre-tests and post-test.

WI target	Word	Translation	IPA	WF target	Word	Translation	IPA
/pʰ-/	pude på	pillow on	[ˈpʰuːð̞̥ə] [ˈpʰɔˑˀ]	/-b̥/	rap	quack	[ˈʁɑb̥]
/tˢ-/	tog tak	train thank you	[ˈtˢɔw] [ˈtˢɑg̊]	/-d̥/	sut	pacifier	[ˈsud̥]
/kʰ-/	ko kage	cow cake	[ˈkʰoˑˀ] [ˈkʰæːæ]	/-g̊/	tak	thank you	[ˈtˢɑg̊]
/b̥-/	bil baby	car baby	[ˈb̥iˑˀl] [ˈb̥ɛjb̥i]				
/d̥-/	dyne dukke	duvet doll	[ˈd̥yːnə] [ˈd̥ɔg̊ə]				
/g̊-/	gynggang *(s)ke	rocking horse sound spoon	[ˈg̊øŋˌg̊ɑŋ] [ˈ(s)g̊eˑˀ]				
/f-/	fugl fut	bird choo	[ˈfuˑˀl] [ˈfud̥]	/-f/	giraf	giraffe	[g̊iˈʁɑf]
/s-/	sove sut	sleep pacifier	[ˈsɒːwə] [ˈsud̥]	/-s/	lys	candle/light	[ˈlyˑˀs]
/ʋ-/	vov varm	woof hot	[ˈʋʌwˀ] [ˈʋɑˑˀm]				
/l-/	lys lege	candle/light play	[ˈlyˑˀs] [ˈlɑːjə]	/-l/	bil	car	[ˈb̥iˑˀl]
				/-ð/	mad	food	[ˈmað̞ˀ]
/j-/	ja	yes	[ˈja]				
/ʁ-/	rap	quack	[ˈʁɑb̥]				
/m-/	mad muh	food mooh	[ˈmað̞ˀ] [ˈmuː]	/-m/	varm	hot	[ˈʋɑˑˀm]
/n-/	nej	no	[ˈnɑjˀ]	/-n/	and	duck	[ˈanˀ]
				/-ŋ/	gynggang	rocking horse sound	[ˈg̊øŋˌg̊ɑŋ]
/h-/	hund	dog	[ˈhunˀ]				

Note: Danish syllable final approximants, except for /ð/, are semivowels and not considered as target consonants. WI= word-initial, WF = word-final. *Young Danish children will typically pronounce the second consonant in adjuncts and the first consonant in true clusters

Appendix 2. The Danish consonant system.

	Bilabial	Labio-dental	Alveolar	Alveo-palatal	Palatal	Velar	Uvular	Pharyngeal	Glottal
Plosives	*p^h b̥		*t^s d̥			*k^h g̊			
Nasals	m		n			**ŋ			
Fricatives		f	s	*ɕ					*h
Glides		*ʋ	**ð̞						
Liquids			l				*ʁ		
Semi-vowels	**w				j			**ɐ̯	

(Basbøll, 2005; Grønnum, 2005) *Only syllable-initial position. **Only syllable-final position.

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Appendix 3. Examples of calculations of the lexical age for NDW.

In the above examples, lexical age scores for NDW are calculated as follows:

5 words at pre-test:	35 words at pre-test:
2.3813ln(5*2) + 9.6897 = 15.17	2.3813ln(35*2) + 9.6897 = 19.81
15 words at post-test:	45 words at post-test:
2.3813ln(15*2) + 9.6897 = 17.79	2.3813ln(45*2) + 9.6897 = 20.41
Gain:	Gain:
17.79 - 15.17 = <u>2.62</u>	20.41 - 19.81 = <u>0.6</u>

Vocabulary growth from five to 15 words corresponds to a 2.62 lexical age gain.

Vocabulary growth from 35 to 45 words corresponds to a 0.6 lexical age gain.

Appendix 4. Intervention program.

Session	Language support strategy	Content
1	Responsive interaction	Observe child's interest, wait with anticipation, respond to child initiative
2	Responsive interaction and expansions/recasting	Imitate, interpret, comment
3	Modelling	Use of target words, aim for 5 productions of the target
4	Speech recasting	What are obstruents, aim for phonological emphasis in target words
5	Books and songs	Incorporate target words into books and/or songs
6	Routines	Incorporate target words in daily routines
7-9	Follow-up	Evaluate parent use of strategies and child progress with parent, introduce new target words

Appendix 5. Observed vocabulary and lexical age at pre-test and post-test.

	CDI				Lexical age CDI				NDW				Lexical age NDW			
	PRE-TEST		POST-TEST		PRE-TEST		POST-TEST		PRE-TEST		POST-TEST		PRE-TEST		POST-TEST	
Pair	INT	CONT	INT	CONT	INT	CONT	INT	CONT	INT	CONT	INT	CONT	INT	CONT	INT	CONT
1	17	10	86	96	16.44	15.17	20.33	20.56	4	4	24	4	14.64	14.64	18.91	14.64
2	25	11	76	80	17.35	15.40	20.00	20.12	4	7	13	25	14.64	15.97	17.45	19.01
3	40	8	132	49	18.47	14.64	21.32	18.96	12	8	42	18	17.26	16.29	20.24	18.22
4	22	13	71	41	17.05	15.80	19.84	18.53	11	3	19	16	17.05	13.96	18.35	17.94
*5**	9	18	66	-	14.92	16.57	19.67	-	2	14	26	40	12.99	17.62	19.10	20.12
6	5	9	83	17	13.52	14.92	20.21	16.44	10	5	35	11	16.82	15.17	19.81	17.05
7	34	21	50	266	18.09	16.94	22.99	19.01	13	8	32	7	17.45	16.29	19.59	15.97
8	6	0	72	30	13.96	10.00	19.87	17.79	5	5	32	16	15.17	15.17	19.59	17.94
9	4	11	45	40	12.99	15.40	18.75	18.47	2	6	7	17	12.99	15.61	15.97	18.09
*10**	0	-	17	5	10.00	-	16.44	13.52	0	5	2	5	10.00	15.17	12.99	15.17
11	20	14	72	26	16.82	15.97	19.87	17.45	9	10	31	39	16.57	16.82	19.52	20.06
12	5	5	45	15	13.52	13.52	18.75	16.14	2	4	31	5	12.99	14.64	19.52	15.17
13	5	4	81	48	13.52	12.99	20.15	18.91	4	5	12	4	14.64	15.17	17.26	14.64
*14**	5	-	34	16	13.52	-	18.09	16.29	4	4	12	7	14.64	14.64	17.26	15.97

*Pair not included in CDI analyses.

Appendix 6. Total consonant inventories (types) at pre-test and post-test.

	PRE-TEST				POST-TEST			
	In all utterances		In words		In all utterances		In words	
Pair	INT	CONT	INT	CONT	INT	CONT	INT	CONT
1	w l j m n ŋ h	w l j m n			g̥ w j m n ŋ h	b̥ d̥ s w l j m n ŋ h	m n h	j
2	j m n ŋ h	w ð j m n ŋ h	m	m h	j m n h	b̥ d̥ w ð j m n h	m h	b̥ g̥ w j m n h
3	b̥ g̥ w j m n h	g̥ w l m n ŋ h	b̥ h	j	b̥ g̥ p^h k^h w j m n ŋ h	b̥ w j m n ŋ h	b̥ d̥ g̥ k^h m n h	m n h
4	g̥ w l j m ŋ h	d̥ g̥ w l j ʁ n ŋ h	h	g̥	d̥ g̥ t^s k^h w ʋ l j m n ŋ h	b̥ d̥ g̥ t^s k^h s x w j m n ŋ h	g̥ h	d̥ g̥ t^s
5	b̥ w j m h	b̥ w l j m n ŋ h		w n h	b̥ d̥ g̥ p^h f w j m n h	b̥ d̥ p^h s x w j m h	b̥ k^h m n h	b̥ p^h f x ʁ m h
6	d̥ g̥ w j m ŋ h	b̥ g̥ w j m n ŋ h	m h	d̥	b̥ g̥ p^h k^h w j m n ŋ h	b̥ d̥ g̥ w j m n ŋ h	g̥ p^h t^s k^h m h	d̥ g̥
7	b̥ g̥ w ð j m n ŋ ʔ Ñ̥	w l j m h	g̥ m	h	g̥ w l j m n ŋ h Ñ̥	b̥ w l j ʁ m n ŋ h	b̥ g̥ m Ñ̥	
8	b̥ g̥ w l j m h	b̥ d̥ w ð l j m h			b̥ d̥ g̥ s w j m n h	b̥ d̥ g̥ s w l j m ŋ h	b̥ d̥ g̥ j m h	g̥ j m h
9	d̥ g̥ w j m ŋ h	l j m n ŋ h		n	b̥ d̥ g̥ t^s k^h s w l j m n h	w ð j m n ŋ h	d̥	w m n h
10	b̥ g̥ s w l j m h	w j m h			b̥ g̥ w j m ŋ h ʔ	w ð l j m h		h
11	w l j m n h	b̥ d̥ w j m n ŋ h	g̥ h	m	g̥ s x w ð l j ʁ m n ŋ h	w l j m n ŋ h	b̥ g̥ m n ŋ h	w j m n h
12	b̥ w j m n h ʔ	b̥ j m h			b̥ d̥ g̥ p^h s w l j m n h	b̥ d̥ g̥ p^h w m h	b̥ d̥ g̥ w m h	b̥
13	w j m ŋ h	b̥ w j m n ŋ h			b̥ d̥ g̥ w j m n ŋ h	b̥ w j m n ŋ h	g̥ m n	
14	w l j m n ŋ h	d̥ w j m h		h	b̥ d̥ w l j m n ŋ h	w ð j m n ŋ h	m	m

STUDY 3

Early intervention in toddlers with cleft palate – Long-term effects on speech accuracy, productive vocabulary, and receptive language

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Abstract

Purpose

This study investigated the long-term effects of parent-implemented focused stimulation intervention in toddlers with cleft palate (CP) and delayed speech and/or language development.

Methods

A quasi-experimental pre-test post-test control group design compared outcomes from 14 toddlers with CP and delayed speech and/or language development who received of parent-implemented focused stimulation (INT group) to a matched control group of 14 toddlers. In addition, speech accuracy results were compared to a group of typically developing (TD) toddlers. All participants attended pre-tests and post-tests after ten months.

Results

Measures of speech accuracy, reported productive vocabulary, and receptive language were not significantly different between the INT group and the CONT group. The INT group was more than three times as likely to not need intervention at post-test as the CONT group, but the difference was not statistically significant. Toddlers with CP had significantly poorer speech accuracy than the TD group, and only a few toddlers with CP performed within two standard deviations of the TD group mean.

Conclusions

Parent-implemented focused stimulation did not affect any of the included speech-language outcome measures six months after intervention. Very few toddlers with CP achieved age-typical speech accuracy levels. Possible explanations for the absence of long-term effects in this study are discussed. Careful consideration of intervention method, dose frequency, and total intervention duration as well as intervention targets is recommended before provision of early intervention to toddlers with CP and delayed speech and/or language development. Future studies of early intervention in toddlers with CP should include longitudinal follow-up.

Introduction

Early speech-language intervention studies in toddlers with CP have not reported results beyond termination of intervention (Broen, Doyle, & Bacon, 1993; Scherer, 1999; Scherer, D'Antonio, & McGahey, 2008; Ha, 2015; Kaiser, Scherer, Frey, & Roberts, 2017; Jørgensen, Scherer, & Willadsen, in preparation). Although there may be potential benefits to intervention effects that are only short-term, e.g. increased or improved parent-child interactions that may in turn increase parent self-efficacy (Trivette, Dunst, & Hamby, 2010), it is important to report long-term follow-up to determine if intervention effects are maintained, and if early intervention has additional long-term effects. Parent-implemented interventions in particular are expected to maintain effects over time because parents are taught to use language support strategies in naturally occurring interactions with their child. This rationale is supported by the longitudinal findings of TD toddlers in Buschmann et al. (2008) and Buschmann, Multhauf, Hasselhorn, and Pietz (2015) who found superior productive language skills in the intervention group at age 3, one year after intervention, and superior receptive language and working memory skills at age 4, compared to a non-treatment control group. Eiserman, Weber, & McCoun (1992) also reported longitudinal follow-up after parent-implemented intervention in toddlers, but compared results to a group of toddlers who received clinician-implemented intervention and found similar effect of parent-implemented and clinician-implemented early intervention. As Eiserman, Weber, & McCoun (1992) did not include a non-treatment control group, it is unknown whether these longitudinal results were a result of intervention or general development. Furthermore, Buschmann and colleagues (2008; 2015) and Eiserman et al. (1992) only targeted language and did not report indirect effects on speech measures.

The present study is an extension of a previous study of early intervention in toddlers with CP and delayed speech and/or language development (Jørgensen et al., in preparation). The previous study reported results from spontaneous speech-language samples and the MacArthur-Bates Communicative Developmental Inventories (CDI: Fenson et al., 2007) and found significant effect on lexical age (a measure of observed productive vocabulary), true consonant inventory in words, and need for intervention immediately after intervention.

Speech accuracy

Speech accuracy is an important outcome measure for children with CP (Allori et al., 2017). Speech accuracy, in particular pertaining to loss of phonemic contrast, is strongly correlated with another important outcome measure; intelligibility (e.g. Kent, Weismer, Kent, & Rosenbek, 1989; Whitehill & Chun, 2002; McLeod, Harrison, & McCormack, 2012; Willadsen & Poulsen, 2012). Intelligibility can be defined as a speaker's ability to convey meaning to a listener which touches upon the fundamental purpose of language (Kent, Miolo, & Bloedel, 1994). For toddlers, intelligibility is not only important for successful communication, but also influences the interaction with caregivers and thereby, indirectly, speech-language development (Tamis-LeMonda, Bornstein, & Baumwell, 2001; Frey, Kaiser, & Scherer, in press). Furthermore, speech accuracy has also been associated with vocabulary size at 30-36 months (Schwarz, Burnham, & Bowey, 2006). In the previous study (Jørgensen et al., in preparation), parents were trained to use speech recasts. Speech recasts are immediate adult responses to meaningful child utterances that add the correct phonological form to the child's utterance. Speech recasts are hypothesised to facilitate learning of new consonants and possibly enhancing speech accuracy by providing immediate comparison between child and adult form in a meaningful context (Fey, Krulik, Loeb, & Proctor-Williams, 1999; Scherer & Kaiser, 2010). Intervention involving speech recasts has been found to affect speech intelligibility positively in language-delayed children older than three years-of-age (Camarata, Yoder, & Camarata, 2006; Yoder, Camarata, & Gardner, 2005). Therefore, a long-term effect on speech accuracy is hypothesised.

Speech accuracy can only be evaluated in meaningful speech as evaluation requires comparison to adult target forms (Stoel-Gammon, 1994). However, evaluation of speech accuracy is a challenge in the early vocabulary as phonological development is closely tied to language level. Children's first word productions are often quite accurate compared to the adult target. This is caused by a selective preference for well-practised sounds, meaning that children produce first words beginning with sounds they already produce in babbling (lexical selectivity; Ferguson & Farwell, 1975; Leonard, Schwartz, Morris, & Chapman, 1981). When a small expressive vocabulary is established, children will typically use their preferable production patterns as phonological templates. These phonological templates may lead to the child's word productions becoming both more similar and less accurate (Velleman & Vihman, 2002). As children learn even more words, it is hypothesized that they begin to recognise similarities across phonological forms (Walley, Metsala, & Garlock, 2003; Stoel-Gammon, 2011). Information about phonemes, phoneme sequences, and syllables is implemented in children's underlying phonological representations to keep the growing number of words distinct (Beckman & Edwards, 2000). This eventually leads to a more systematic phonology where more consistent phonological processes can be observed (Ferguson & Farwell, 1975; Vihman, 2014). Thus, speech accuracy will depend on language level.

Speech samples. Speech accuracy is affected by speech samples (Stoel-Gammon & Williams, 2013). In spontaneous speech samples, some children may attempt simple one-syllable words with relative success while others attempt more complex multi-syllabic words resulting in less accurate productions. This difference may result in higher speech accuracy scores in children whose phonological development is actually less advanced than in children with lower accuracy scores. Accordingly, standardised naming tests are appropriate when comparing speech accuracy across children.

Profiles of Early Expressive Phonological Skills. Most phonological naming tests are not suitable for toddlers because they involve words that are outside a toddler's vocabulary, and because words are elicited from pictures. Naming from pictured stimuli is a difficult task for many toddlers (Stoel-Gammon & Williams, 2013). Profiles of Early Expressive Phonological Skills (PEEPS) was developed as a test of phonological development in two-year-olds with typical development (TD) and older children with delayed vocabulary acquisition (Stoel-Gammon & Williams, 2013). The PEEPS test uses objects instead of pictures and is aimed at assessing speech from age two. Test items were selected based on age-of-acquisition (AoA) data from the American CDI norms (Fenson et al., 2007) and included words produced by at least 50 % of toddlers at 24-30 months. Further, words were selected to elicit target consonants across all places, voice, and manner categories in English as well as in different syllable structures. Stoel-Gammon and Williams (2013) tested 40 TD English-learning toddlers and 17 toddlers with CP aged 17-37 months with the PEEPS test. At 23-25 months, the TD toddlers on average named 91% of the target words whereas the toddlers with CP named 73%. A Danish version of the PEEPS test, Test af Ekspressive Fonologiske Færdigheder (TEFF, *Eng.* Test of expressive phonological abilities) was tested on 98 TD Danish-learning toddlers aged 24-35 months (Holmer and Pedersen, 2017). In TEFF, target words were selected based on AoA data from the Danish CDI norms (Wehberg et al., 2007) that were produced by at least 70% of toddlers at 24 months. It involves 55 test items and includes three examples of all Danish word-initial phonemes and two examples of all Danish word-final phonemes. The Danish toddlers named 99% of the target words on average.

The toddlers with CP in the previous study (Jørgensen et al., in preparation) were included based on their small reported productive vocabularies (below 50 words and/or below the age-equivalent 10th percentile on the MacArthur-Bates Communicative Developmental Inventories (CDI: Fenson et al., 2007) and a speech criterion (absence of alveolar stops). They were too young to participate in a naming test, and most of them produced only a few words during pre-testing, and therefore, analysis of speech accuracy was not performed.

Measures of speech accuracy

Percent consonants correct – adjusted (PCC-A). Shriberg and Kwiatkowski (1982) developed percent consonants correct (PCC) as a severity index for disordered speech. PCC measures the proportion of consonants produced correctly based on phonetic transcription. Shriberg, Austin, Lewis, McSweeny, and Wilson (1997) proposed PCC-A to differentiate between common distortion errors and uncommon or idiosyncratic distortion errors. PCC was originally developed for spontaneous speech, but is now widely used as a measure of correctness in naming tests. When PCC is used with naming tests, benchmarks for impairment severity are not appropriate (Shriberg et al., 1997). Different measures of PCC have been reported as outcome measures in studies of early intervention in toddlers with CP (Scherer, 1999; Scherer et al., 2008; Kaiser et al., 2017). The International Consortium of Health Outcome Measures very recently recommended outcome measures that reflect most important outcomes for patients with CP, including use of PCC for speech evaluation in children with CP (Allori et al., 2017).

The Weighted Measure of Speech Sound Accuracy (WSSA). The WSSA was developed and evaluated by Preston, Ramsdell, Oller, Edwards, and Tobin (2011) based on the Weighted Reliability Measure for Phonetic Transcription (Oller & Ramsdell, 2006) in an effort to quantify speech sound accuracy. Where PCC only counts consonants as correct or incorrect, the WSSA assigns weight to each error, based on widely recognised tenets of phonological theory (Oller & Ramsdell, 2006). Furthermore, the WSSA contains both a global structural level and a featural level. For each word, the actual production is aligned with and compared to the target production. Each level attains a score between 0.00 and 1.00 based on agreement of matching nuclei and consonantal segments (global structural level) and the agreement of phonetic features of vowels and consonants (featural level). The scores from each level are multiplied to compute the total score. Phoneme deletions are weighted most on the global structural level, e.g. 'cup' produced as [ˈʌp] yields a lower score than [ˈtʌt]. Major changes in manner, placement, or voicing are weighted most on the featural level, e.g. 'cup' produced as [ˈweŋ] yields a lower score than [ˈtʌt]. Both examples would share the same PCC-A score whereas WSSA scores would be different. Examples of WSSA calculations are shown in appendix 1. Preston et al. (2011) found that WSSA separated children with and without speech sound disorders, and that WSSA was superior to PCC in capturing speech accuracy growth over time in toddlers. Further, Preston et al. (2011) suggested that WSSA may be sensitive to atypical speech errors. Hence, the WSSA is potentially a useful tool for quantifying speech accuracy beyond PCC in toddlers with CP and delayed speech and/or language development.

Language measures

Reported productive vocabulary growth is slower in toddlers with CP between 14-21 months compared to toddlers without CP. This difference is explained by a steeper vocabulary acquisition rate in toddlers without CP (Broen, Devers, Doyle, Prouty, & Moller, 1998; Lu, Ma, Luo, & Fletcher, 2010; Hardin-Jones & Chapman, 2014; Willadsen, 2012). Productive vocabulary was targeted alongside consonant inventory in five of six earlier studies of early intervention in toddlers with CP (Scherer, 1999; Scherer et al., 2008; Ha, 2015; Kaiser et al., 2017, Jørgensen et al., in preparation). Studies including control groups of toddlers with CP reported increased reported productive vocabulary (Ha, 2015; Kaiser et al., 2017), total number of observed words (Ha 2015), and lexical age (an age-equivalent score developed to capture productive vocabulary development; Jørgensen et al., in preparation), but none have reported longitudinal outcomes. Investigations of receptive language development in toddlers with CP have mostly found that toddlers with CP perform within normal limits on receptive language tests, although poorer compared to peers without CP (Long & Dalston, 1983; Scherer & D'Antonio, 1995; Snyder & Scherer, 2004; Lu et al., 2010; Broen et al., 1998). However, a few studies found delayed receptive language skills compared to standardised norms (Jocelyn, Penko, & Rode, 1996; Lamônica, Silva-Mori, Ribeiro, & Maximino, 2016). Of particular concern is that the delay seemed to increase from 12-24 months in the longitudinal study by Jocelyn et al. (1996). Naturalistic speech-language interventions are based on a transactional perspective of language acquisition where parents are taught to be responsive to their child's utterances and respond with contingent language (Tannock & Girolametto, 1992). This may not only aid child productive language, but may also help children realise how objects or actions in a natural, meaningful context are related to language form (Brinton & Fujiki, 1995). However, early intervention has in general yielded a low effect on receptive language (Law, Garret, & Nye, 2004). Receptive language has not been directly targeted in earlier studies of early intervention in toddlers with CP, but Kaiser et al. (2017) did report that early intervention affected receptive language when compared to a control group of toddlers with CP.

Research questions

This study investigated the long-term effects of parent-implemented focused stimulation (FS) in toddlers with CP and delayed speech and/or language development compared to a matched control group of toddlers with CP. The study addressed the following questions:

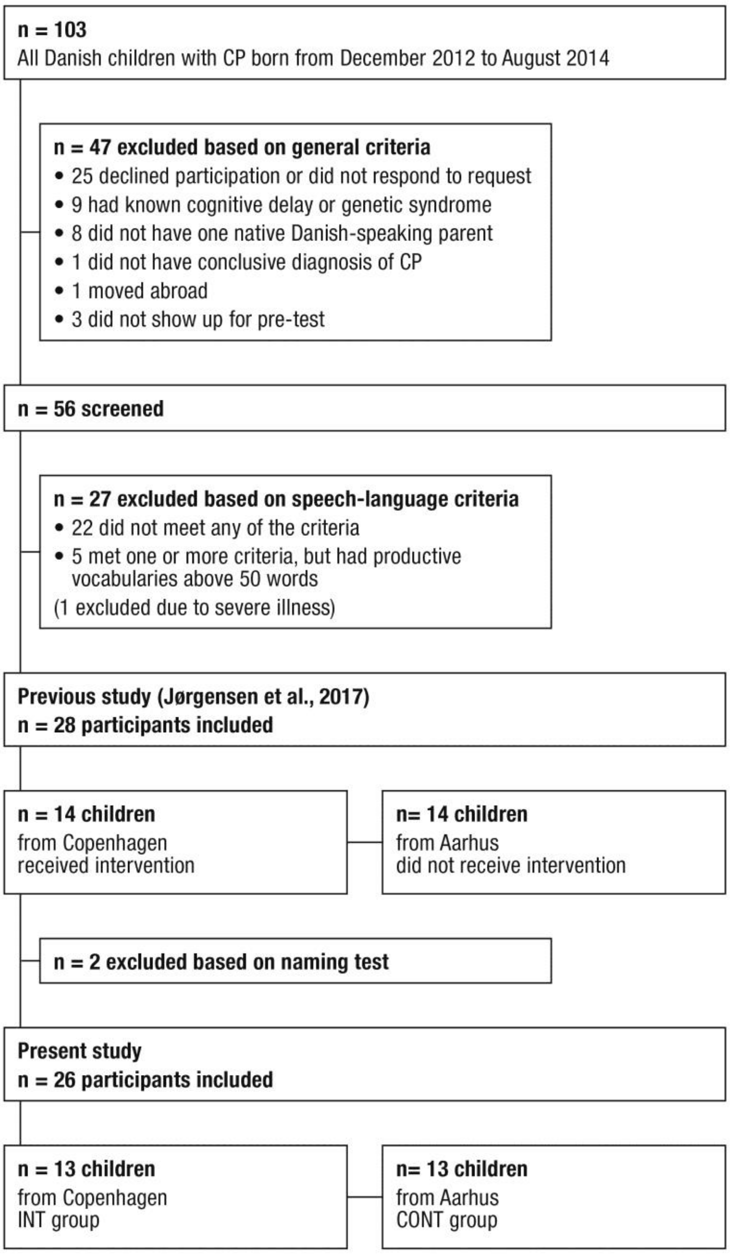
1. Does FS have long-term effect on PCC-A and WSSA in a naming test?
2. How does speech PCC-A and WSSA in the two groups of toddlers with CP compare to TD Danish toddlers without CP?
3. Does FS have long-term effect on reported productive vocabulary and receptive language?
4. Does FS affect continued need for intervention?

Method

Participants

The participants were 26 toddlers with CP aged 25-35 months. Six months prior to the present study, fourteen toddlers had received nine sessions of home-based parent-implemented FS intervention for three months (INT group). Fourteen toddlers did not receive parent-implemented FS (CONT group) but were allowed to seek local speech therapy services. Two toddlers (one from the INT group and one from the CONT group) were excluded from the present study because they only named few words (six and 12 of 55 target words). The eligibility procedure is shown in figure 1.

Figure 1. Flowchart of the eligibility procedure.



Participants were recruited from a complete birth cohort of all 103 Danish toddlers with CP born between December 2012 and August 2014. They were included based on general criteria: cleft palate ± cleft lip, at least one native Danish-speaking parent, and absence of a known genetic syndrome, severe cognitive delay, or sensorineural hearing loss, and speech-language criteria: reported vocabulary below 50 words and at or below the 10th percentile on the Danish version of the CDI (Andersen, Vach, Wehberg, & Bleses, 2006), and/or no alveolar stops. Information was based on parental report and review of medical records. Speech-language criteria were determined from combined information from the CDI and real-time listening (Ramsdell, Oller, Buder, Ethington, & Chorna, 2012). The intervention program with details about the inclusion criteria and the screening procedure is described in Jørgensen & Willadsen (2017) and Jørgensen et al. (in preparation).

There were no statistically significant differences between groups at pre-test for reported vocabulary size, reported vocabulary size for age, age, age at palatal surgery, cleft type, or socio-economic status[7] (SES), but there was a significant group difference in time from palatal surgery to pre-test (see table 1). Participants in the INT group had on average had a complete palate for two months longer than the CONT group. CDI questionnaires were missing for two CONT participants at pre-test, but were available from post-test. Therefore, statistical analyses were run again with these participants' scores set at both zero and post-test scores which still revealed no statistically significant group differences. At pre-test, all participants performed within the "competent" or "emerging" categories of the Bayley 3 screening subtests of cognitive and receptive language skills (see Jørgensen et al., in preparation). At post-test, there was also no significant difference found for age.

Participant characteristics are shown in table 2, including history of otitis media with effusion (OME) and history of pressure-equalizing tubes (PETs) before 12 months and between 12-18 months. Results from hearing tests were not available.

Written consent to participation was obtained from all parents, complying with ethical principles of the Declaration of Helsinki. The project is registered with the Danish Data Protection Agency.

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[7] SES is based on highest level of education and current employment status (Ploug, Andersen, Olsen, & Juul, 2012).

Table 1. Between-group comparison of matching variables at pre-test and post-test six months after intervention.

	INT group			CONT group			95% BCa CI	*p*-value
	M (median)	SD	Min-max	M (median)	SD	Min-max		
CDI productive vocabulary at pre-test	15.15 (9)	12.25	4-40	10.82 (11)	5.88	0-21	[-2.82-12.27]	.273
CDI percentile at pre-test	8.46 (5)	8.20	0-25	7.27 (5)	9.18	0-30	[-6.13-7.86]	.741
Age at pre-test	19.31 (20)	2.29	17-24	18.36 (19)	1.29	16-20	[-0.57-2.48]	.238
Age at post-test	29.62 (28)	2.69	27-35	28.15 (28)	1.68	25-31	[-0.28-3.33]	.110
Age at palatal surgery	13.54 (14)	1.13	12-16	14.00 (14)	1.28	12-16	[-1.39-0.50]	.347
Time from palatal surgery to pre-test	5.92 (6)	2.06	4-10	4.00 (4)	1.48	2-7	[0.50-3.42]	.021*
Cleft type+	---	---	---	---	---	---	---	.156
SES++	3.00 (3)	0.91	2-4	3.55 (4)	1.04	2-5	---	.139

*p< 0.05. + Pearson's chi-squared test, ++Wilcoxon rank sum test.

Table 2. Participant characteristics.

INT group							CONT group						
Child	Gender	Cleft type	Age in months	SES	History of OME	PETs before 12 m/ PETs 12-18 m	Child	Gender	Cleft type	Age in months	SES	History of OME	PETs before 12 m/ PETs 12-18 m
1	M	UCLP	35	4	y	n/n	1	M	BCLP	30	3	y	uk/y
2	M	UCLP	34	2	y	y/y (one ear)	2	F	CL/SUBM	31	3	y	n/n
3	M	UCLP	30	4	y	n/y	3	M	UCLP	27	3	uk	n/n
4	M	CPO	30	3	y	n/y	4	M	CPO	29	2	y	y/n
5	M	BCLP	28	3	y	y/n	5	F	UCLP	30	4	y	n/y
6	F	BCLP	27	2	y	y/y	6	F	CPO	28	5	y	n/y
7	M	UCLP	32	4	y	y/y	7	F	CPO	28	4	y	n/y
8	M	UCLP	28	4	y	y/y	8	M	UCLP	28	4	y	y/y
9	M	UCLP	27	2	y	y/y	9	F	CPO	28	5	y	y/n
							10	M	UCLP	27	4	y	y/y
11	M	UCLP	31	4	y	n/y	11	M	UCLP	29	2	y	y/y
12	M	UCLP	28	2	y	n/y							
13	M	UCLP	27	2	y	n/y	13	M	UCLP	26	4	y	n/y
14	M	CPO	27	3	y	y/y	14	M	UCLP	25	4	y	y/n
Mean			29.5	3.00						28.2	3.62		

*OME treated with hearing aids from 5-25 months.

M=male, F=female, UCLP=unilateral cleft lip and palate, CPO=cleft palate only, BCLP=bilateral cleft lip and palate, CL/SMCL=cleft lip and submucous cleft palate, y=yes, n=no, uk=unknown.

Design and procedures

Prior to testing, parents completed the CDI. A naming test was administered and video recorded in the cleft clinic by two different SLPs. The naming test was TEFF (Holmer & Pedersen, 2017), the Danish version of the PEEPS test (Stoel-Gammon & Williams, 2013). The test items were packed in five separate bags organized in play themes. One bag at the time was presented to the child who was allowed to grab any item of interest from the bag. The SLP encouraged spontaneous naming by expectant waiting and only asked 'what is it?' if spontaneous naming did not occur. Then, the SLP added semantic cues, prompted by asking 'I think it is a dog, what do you think it is?' before finally asking for repetition. If an item was not named, it was put aside and introduced again later. A list of test items is found in appendix 2.

Transcription and reliability

Video recordings of the naming test were segmented into separate video clips of the individual test items in Phon (Rose & MacWhinney, 2014). If a child produced more than one version of a test item, spontaneous naming was preferred before first version. Furthermore, if participants did not name a test item, attempts were made to find phonetically similar words produced by the participant in the recording, e.g. word-initial /d/ in 'dør' ([ˈd̥œʌ̯ˀ], Eng. 'door') would be replaced with 'der' ([ˈd̥eʌ̯ˀ], Eng. 'there'), rather than 'dame' ([ˈd̥æːmə], Eng. 'lady'). Test items were transcribed in semi-narrow IPA. An SLP with 15 years of experience in transcribing CP speech, who was blinded to group allocation, transcribed the entire material. A second SLP (the author) with 12 years of experience in transcribing CP speech transcribed 25% of the material from each participant for inter-rater evaluation. For intra-rater evaluation, both raters re-transcribed 25% of the material from each participant. The weighted reliability measure (WRM) (Oller and Ramsdell, 2006) was used for evaluation of consonant inventories. Inter-rater WRM was .85, and intra-rater WRM was .97 and .91, respectively. Vowel quality was not evaluated in the present study which may in part explain the high agreement values.

Outcome measures

Percent consonants correct – adjusted (PCC-A). PCC-A was calculated for all consonants in target words, except word final /w j/ which are considered semi-vowels in Danish. In addition, PCC-A for word-initial (WI) consonants was calculated separately because the intervention involved word-initial target consonants in target words. PCC was adjusted by excluding the following errors in the analyses: [s] realised as [θ ɕ ɬ ç] was not counted as an error because these distortions are common in Danish-speaking pre-schoolers (Hutters & Bau, 2006). Minor articulatory errors such as bilabial realisation of labio-dentals and vice versa and palatal realisation of velars were also not counted as errors. Finally, nasal emission on high pressure consonants was disregarded. The participants were too young to cooperate with the naming test at pre-test; hence, PCC-A gain scores could not be calculated. Since no statistically significant differences between groups were found at pre-test for any measure of vocabulary or consonant inventory (see Jørgensen et al., in preparation), it was assumed that groups would also not differ on speech accuracy. The outcome measure was difference in total PCC-A scores and WI PCC-A scores between the INT group and the CONT group. The INT group had gained more word-initial true consonant types in words than the CONT group immediately after intervention. This result was statistically significant with a large effect size. Also, a large effect size indicated that the INT group had gained more word-initial oral stop tokens in words than the CONT group immediately after intervention (Jørgensen et al., in preparation). Therefore, the INT group may have been practising true consonants and oral stops to a greater extent than the CONT group, possibly leading to more correct productions of these consonants in the naming test six months after intervention. Thus, between-group differences in total and WI PCC-A of true consonants and oral stops were investigated.

WSSA. WSSA was calculated based on whole-word transcription of all target words. Vowels were included in the global structural analyses, but not in the featural analyses where vowels were counted as correct unless they substituted a consonantal segment. The outcome measure was difference in WSSA between the INT group and the CONT group.

Comparison of PCC-A and WSSA between toddlers with and without CP. PCC-A and WSSA scores were available from 98 TD toddlers (Holmer & Pedersen, 2017). Outcome measures were group differences in PCC-A and WSSA between the two groups of toddlers with CP and the TD group.

Language measures

Reported productive vocabulary. Reported productive vocabulary was obtained from CDI questionnaires. The outcome measure was between-group difference in CDI productive vocabulary scores.

Receptive language. The New Reynell Developmental Language Scales (NRDLS – Danish version: Edwards, Letts, & Sinka, 2015) was used to test participants' receptive language levels. The outcome measure was between-group difference in NRDLS standard scores.

Continued need for intervention. This measure compared how many participants met the pre-test speech-language inclusion criteria in each group. In the previous study (Jørgensen et al., under review), the language inclusion criterion (at or below the 10th percentile for reported productive vocabulary) was derived from CDI questionnaires, and the speech inclusion criterion (no alveolar stops) was derived from consonant inventories in spontaneous utterances (both meaningful utterances and babbling). Whereas CDI questionnaires were also available in the present study, spontaneous speech was not. As participants had grown older, and the speech sample in the present study was a naming test, it was a concern that absent/present alveolar stops would be too crude a measure to capture a difference between groups. Chapman (2004) reported that correlations were strongest between emerging speech skills in children with CP at 21 and 39 months, and Holmer and Pedersen (2017) found that emerging consonants (at least 50% of consonants produced correctly) separated Danish toddlers without CP into younger and older age groups. Therefore, the speech criterion was changed from absence of alveolar stops to non-emerging alveolar stops, defined as less than 50% correctly produced alveolar stops. The outcome measure was between-group difference in continued need for intervention at post-test.

Statistics

Statistical analyses were performed using SPSS statistical package version 22 (SPSS Inc., Chicago, IL). Robust independent t-tests with bias-corrected and accelerated bootstrapping (BCa) were used in between-group analyses. Mann Whitney U tests were used in cases where BCa could not be performed. Effect sizes are reported along with p-values in this study to supplement analyses of statistical significance with clinical magnitude. Interpretations of effect sizes and reliability measures are based on commonly cited benchmarks (Cohen's *d*: .2 small, .5 medium, .8 large (Cohen, 1988)).

Results

PCC-A scores

Mann-Whitney U-tests compared the number of words produced in the naming test and the total number of consonants produced between groups. There was no significant difference for these measures between the INT group (mean no. of words: 46.69; mean no. of consonants: 71.54) and the CONT group (mean no. of words: 47.15; mean no. of consonants: 72.62), p=.898 and p=.719.

PCC-A for all WI consonants, WI true consonants, and WI oral stops were compared between groups by independent t-tests with BCa (see table 3). The INT group had the highest score for all tree measures. However, none of the differences were statistically significant, and effect sizes were small.

PCC-A for all consonants, true consonants, and oral stops were compared between groups by independent t-tests with BCa. The INT group had the highest score for oral stops. As shown in table 3, none of these differences were statistically significant, and effect sizes were small.

Table 3. Comparison of PCC-A (all measures) between the INT group and the CONT group.

PCC-A	INT group			CONT group			*p*	95% BCa CI	*d*
	M (median)	SD	min-max	M (median)	SD	min-max			
All WI consonants	0.49 (0.46)	0.21	0.17-0.91	0.44 (0.44)	0.17	0.12-0.66	.542	[-0.11-0.20]	0.23
WI true consonants	0.49 (0.52)	0.26	0.10-0.93	0.43 (0.39)	0.20	0.11-0.74	.507	[-0.12-0.24]	0.26
WI oral stops	0.48 (0.56)	0.32	0.00-0.90	0.36 (0.33)	0.25	0.00-0.94	.317	[-0.13-0.34]	0.41
All consonants	0.44 (0.40)	0.22	0.14-0.87	0.45 (0.47)	0.18	0.05-0.70	.897	[-0.17-0.16]	-0.05
True consonants	0.43 (0.39)	0.23	0.10-0.86	0.44 (0.48)	0.19	0.04-0.74	.916	[-0.16-0.18]	-0.04
Oral stops	0.48 (0.44)	0.29	0.00-0.92	0.40 (0.42)	0.25	0.00-0.91	.465	[-0.16-0.31]	0.29

WSSA

An independent t-test with BCa compared WSSA scores between groups and revealed no statistically significant group difference (mean WSSA: 0.79 in both groups), t(24)=0.019, p=.978, 95% BCa CI [-0.09-0.09], *d*=0.01.

Comparison with TD toddlers

Since differences in PCC-A and WSSA scores between the INT and the CONT group were not significant, results from the INT group and the CONT group were pooled in comparisons with the TD toddlers. There was no statistically significant age difference between the toddlers with CP (mean age: 28.88; min-max: 25-35 months) and the TD group (mean age: 29.78, min-max: 24-35 months), p=.120.

Toddlers with CP produced fewer words (mean: 46.92) than the TD toddlers (mean: 54.30). An independent t-test with BCa revealed that this difference was statistically significant with a large effect size, t(25.505)=5.927, p=.001, 95% BCa CI [4.80-9.84], *d*=2.33.

As shown in table 4, the toddlers with CP had lower PCC-A and WSSA scores than the TD toddlers. These differences were statistically significant with large effect sizes.

Table 4. Comparison of PCC-A and WSSA between toddlers with CP and TD toddlers.

	Toddlers with CP			TD toddlers			*p*	95% BCa CI	*d*
	M (median)	SD	min-max	M (median)	SD	min-max			
PCC-A	0.44 (0.45)	0.20	0.05-0.87	0.88 (0.91)	0.10	0.52-1.00	.001*	[0.37-0.51]	2.42¤
WSSA	0.80 (0.81)	0.09	0.58-0.96	0.97 (0.98)	0.02	0.88-1.00	.001*	[0.14-0.21]	2.02¤

**p*= 0.001, ¤= large effect size.

Language measures

Reported productive vocabulary. The INT group had higher CDI productive vocabulary scores (mean: 285 words) than the CONT group (mean: 254 words), but the difference was not significant with a small effect size, t(22)=.765, p=.452, 95% BCa CI [-55.73-137.64], d=0.3.

Receptive language. The INT group had higher NRDLS standard scores (mean: 91.85) than the CONT group (mean: 83.69). An independent t-test with BCa revealed the difference was not significant with a medium effect size: t(24)=1.607, p=.121, 95% BCa CI [-2.00-17.22], *d*=0.63.

Continued need for intervention. Eight/13 toddlers in the INT group and 11/13 toddlers in the CONT group still matched the criteria for need for intervention. This difference was not significant, p=0.378. Based on the odds ratio, the INT group was 3.4 times more likely not to need intervention at post-test.

All PCC-A, WSSA, CDI, and NRDLS standard scores are shown in appendix 3.

Discussion

Early intervention studies in toddlers with CP are few and far between, and no study to this date has investigated the long-term effects of early intervention in toddlers with CP. Positive long-term effects would provide justification for spending societies' limited resources on early intervention and adding to families' burden of care. The present study investigated the long-term effect of parent-implemented FS in toddlers with CP and delayed speech and/or language development. Results were compared to a matched control group of toddlers with CP.

PCC-A and WSSA

The INT group had higher PCC-A scores than the CONT group for all WI consonant measures, and for PCC-A of oral stops. However, none of the differences were statistically significant, and effect sizes were small. In addition, there was no statistically significant difference in WSSA between the INT and the CONT group. This result indicates that the two groups produced similar amounts of errors as well as error types of similar weight. Preston et al. (2011) found that WSSA was superior to PCC-A in capturing phonological development in toddlers over time. This indicates that WSSA would be useful for measuring intervention effect on phonological development. In this study, however, pre-test WSSA could not be calculated because at pre-test, the participants were too young to comply with a phonological naming test. Therefore, analysis of WSSA over time could not be performed. Even though the present study targeted both vocabulary and consonant inventory directly, results are in line with the studies of Girolametto, Pearce, and Weitzman (1997) and Whitehurst et al. (1991). These authors reported that early intervention with specific word targets but only within-inventory consonants did not affect gain in PCC.

There are a number of possible explanations for the lack of effect on PCC-A. First of all, the participants in the present study generally had low language levels. This may have affected parents' frequency of speech recasting because use of more meaningful utterances may provide parents with more opportunities for speech recasting. Speech recasting is assumed to facilitate PCC-A because the speech recast provides an immediately contrast between child production and adult target. At the end of the intervention program, three of 11 parents did not meet the criterion level for expansions including speech recasting (Jørgensen et al., 2017, in preparation). However, parents' use of speech recasts was not measured beyond this time point. Further, as the intervention method in this study involved more components than speech recasting, it is not possible to determine the particular effect of speech recasts. Another explanation related to low language levels is that the task of naming words may have been so cognitively taxing for participants that speech accuracy levels decreased compared to spontaneous word productions. In addition, participants may not all have developed systematic phonologies yet. In both cases, it may have been more informative to investigate consonant inventories and speech accuracy in spontaneous word productions. However, spontaneous speech samples would have introduced difficulties in comparing participants because participants would not have produced the same words and speech accuracy may have been affected by different word selection strategies.

Finally, toddlers with CP have restricted consonant inventories and phonological delay that are most often the consequences of the early structural deficit. Therefore, they may need more time to practice than toddlers without CP before intervention effects can be achieved or become evident, especially concerning stops. Extending intervention with more training or follow-up sessions may be necessary. Along the same line of reasoning, intervention methods that require production of intervention targets, such as enhanced milieu teaching (EMT), may be more beneficial for increasing speech accuracy in toddlers with CP because production practice may be enhanced in methods like EMT. Kaiser et al. (2017) employed clinician-implemented EMT intervention in a study of toddlers with CP who received two weekly sessions over six months and reported a short-term effect on PCC-A compared to a BAU control group of toddlers with CP. Even though results were reported immediately after intervention, the cumulative intervention intensity (dose*dose frequency *total intervention duration; Warren, Fey, & Yoder, 2007) may have yielded more target word productions from the toddlers in Kaiser et al. (2017) during the entire period of intervention compared to the present study. However, interpretation of the results of Kaiser et al. (2017) should be cautioned because participants differed on mean pre-test PCC-A levels.

Comparison between the CP group and the TD group

Statistically significant differences with large effect sizes were found between the toddlers with CP and the TD group for both PCC-A and WSSA. Follow-up analyses showed that ten CP toddlers and 40 TD toddlers had PCC-A scores between the lowest scoring TD toddler and the highest scoring CP toddler (figure 2), and seven CP toddlers and 30 TD toddlers had WSSA scores between the lowest scoring TD toddler and the highest scoring CP toddler (figure 3). Thus, WSSA may be slightly superior in differentiating typical and delayed/disordered speech.

Figure 2. Distribution of PCC-A scores in the CP group and the TD group. Reference lines represent the lowest scoring TD toddler and the highest scoring CP toddler (0.52-0.87).

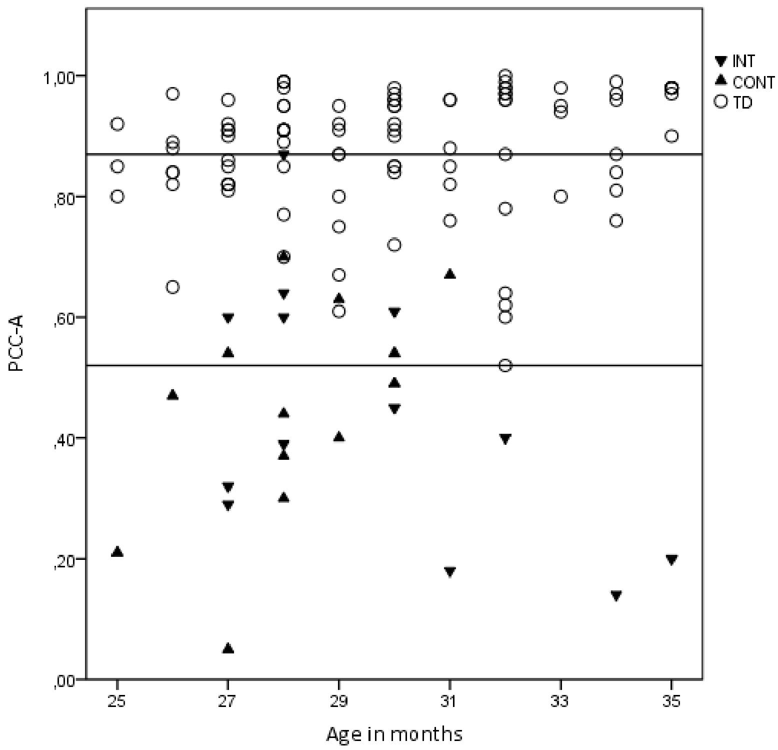
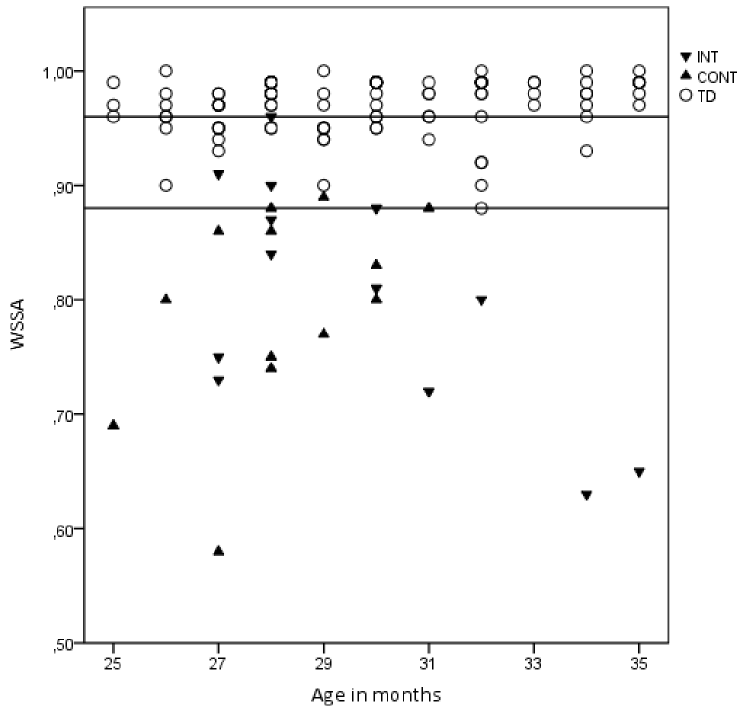


Figure 3. Distribution of WSSA scores in the CP group and the TD group. Reference lines represent the lowest scoring TD toddler and the highest scoring CP toddler (0.88-0.96).



It is likely that some of the 98 TD toddlers had a phonological delay or disorder. Hence, follow-up analyses were performed with reference lines for both PCC-A and WSSA set at two standard deviations (SDs) below the mean for TD toddlers. Only two toddlers with CP had PCC-A scores above two SDs from the mean PCC-A score of the TD group, and only one toddler with CP had a WSSA score above two SDs from the mean WSSA score of the TD group (see figures 4 and 5). Seven and six TD toddlers, respectively, scored below two SDs of the mean corresponding to 5.9% and 4.9% of all 98 TD toddlers. These percentages are close to reported estimates of speech delay although estimates vary considerably (see Shriberg, Tomblin, & McSweeny, 1999; and Law, Boyle, Harris, Harkness, & Nye). Hence, it seems that very few toddlers with CP had obtained age-typical speech accuracy.

Figure 4. Distribution of PCC-A scores in the CP group and the TD group. Reference lines are set at 2 SDs below the mean of the TD toddlers (0.68).

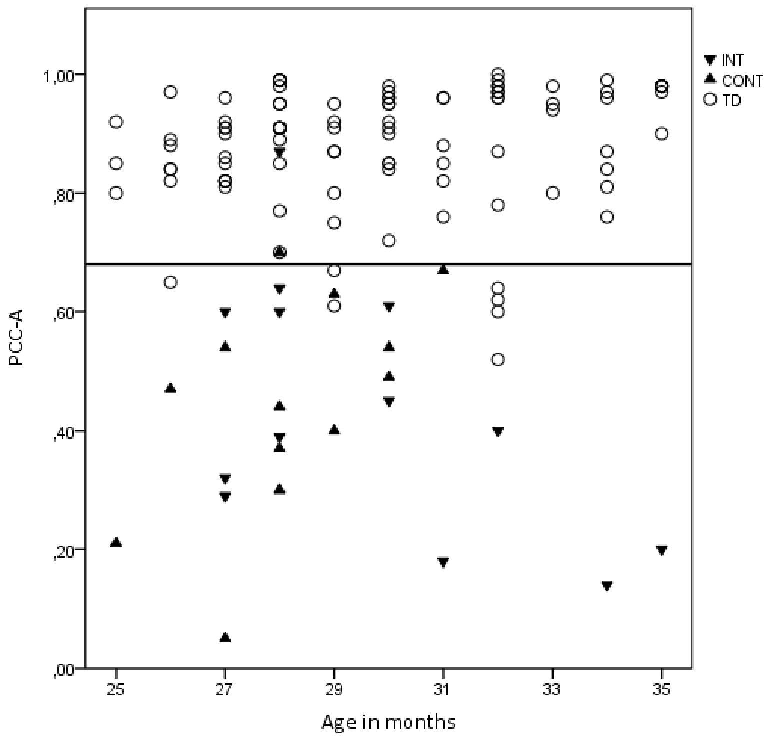
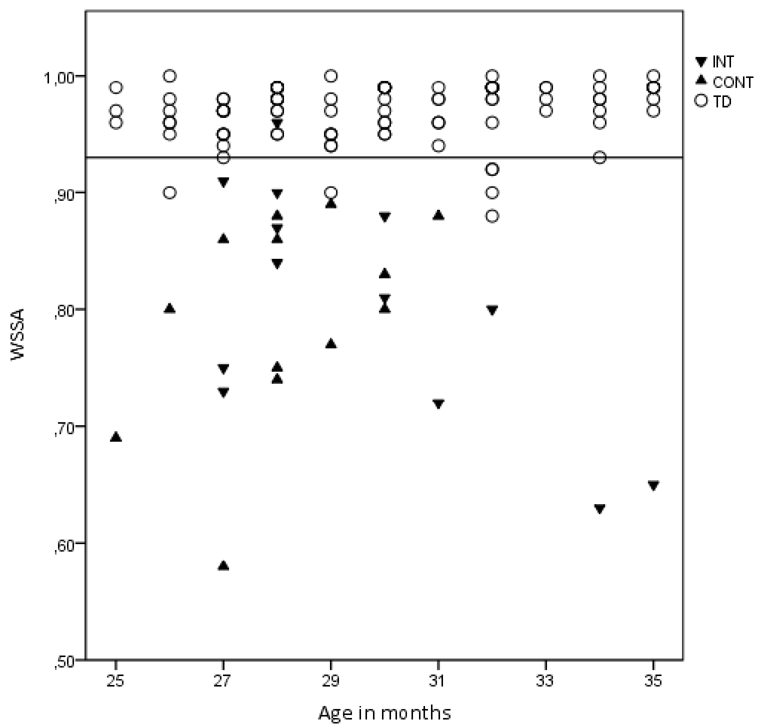


Figure 5. Distribution of WSSA scores in the CP group and the TD group. Reference lines are set at 2 SDs below the mean of the TD toddlers (0.93).



Scherer, Williams, Stoel-Gammon, and Kaiser (2012) compared PCC scores from the PEEPS test in 24 toddlers with CP aged 18-36 months to 42 TD peers. The mean PCC score of the TD toddlers was highly similar to the TD group in the present study (86.5% vs. 88%) whereas the PCC-A scores of the CP group were lower in the study by Scherer et al. (2012) (34.9% vs. 44% in the present study). The PCC-A scores from the present study and the PCC scores from Scherer et al. (2012) cannot be compared directly. First of all, PCC-A is likely to yield a higher score than PCC because it does not count common distortions as errors. Secondly, differences in age, inclusion of clusters, and, not least, languages make comparison difficult. However, both English- and Danish-learning toddlers with CP had significantly poorer speech accuracy than their TD peers.

The CP group in the present study named significantly fewer words than the TD group, even though efforts were made to substitute target words with similar words produced by the CP group. This group difference may reflect differences in language levels. The pre-test inclusion criteria for the CP group were reported productive vocabulary below 50 words and/or below the 10th percentile on the CDI. At the time of the present study, eleven/26 toddlers with CP had reported productive vocabularies at or below the 10th percentile on the CDI, and eight/26 toddlers scored below the 10th percentile on the receptive language subtest of the NRDLS (three/26 scored below 2 standard deviations on the NRDLS, see appendix 3). Although the TD group was not language tested, it is likely that they would have performed better than the toddlers with CP. Stoel-Gammon & Williams (2013) also found that toddlers with CP produced less target words and consonants compared to TD toddlers although this difference decreased gradually until age 3 where almost no difference was found. The language levels of the participants in Stoel-Gammon & Williams (2013) are not reported. Albeit toddlers with CP may not be able to name as many words on a phonological naming test as TD toddlers, TEFF still provided relatively large speech samples from toddlers with CP. This indicates that PEEPS/TEFF is a viable option for testing speech accuracy in toddlers with CP.

Language measures

The INT group had higher CDI productive vocabulary scores than the CONT group, but the difference was not statistically significant with a small effect size. In the previous study (Jørgensen et al., in preparation), a measure of lexical age was constructed to account for exponential productive vocabulary growth from 0-200 words. However, reported productive vocabulary growth is not exponential from 200-600 words (Andersen et al., 2006). At pre-test, CDI productive vocabulary scores ranged from 0-40, but at post-test they ranged from 5-576 words. Thus, neither gain in raw CDI scores or lexical age scores would have reflected individual vocabulary growth trajectories. Instead, it may be more productive to compare how many toddlers in each group scored at or below the 10th percentile on the CDI at pre-test, but above the 10th percentile at post-test. Six toddlers in each group fit this description thus still not revealing any group difference.

The INT group had higher receptive language scores than the CONT group, but the difference was not statistically significant with a medium effect size. Law et al. (2004) found that early intervention in general has shown low effect on receptive language. However, Roberts and Kaiser (2011) reported that effect on receptive language is greater in clinician-implemented interventions which may suggest that parent-implemented intervention may not be appropriate when receptive language is the primary intervention goal. Nonetheless, this result of the present study should be interpreted with caution since participants' receptive language levels were evaluated with a screening test at pre-test and thus it cannot be ruled out that pre-test differences existed. In addition, many toddlers found it difficult to cooperate with the NRDLS which limits the interpretation of results.

Kaiser et al. (2017) reported intervention effect on both reported productive vocabulary and receptive language in their study of toddlers with CP, but again, the interpretation of results from Kaiser et al. (2017) must be cautioned because the intervention group and the BAU group differed in mean pre-test reported productive vocabulary and receptive language levels.

Continued need for intervention

The INT group were 3.4 times more likely to not need intervention than the CONT group at post-test, but the difference between groups was not statistically significant. In contrast, need for intervention was significantly greater in the CONT group immediately after intervention (Jørgensen et al., in preparation).

These results suggest that the intervention program accelerated speech-language development, but that rate of development did not continue at group level after the program ended. As seen in table 5, the CONT group seemed to catch up on language development to some extent, but not speech development. Continued development may possibly be aided by extending the intervention program with more parent follow-up sessions.

Table 5. Participant need for intervention across the intervention study in both groups.

			Pre-test	After 4 months	After 10 months
INT group	Need for intervention	- No alveolar stops/	9	4	7
		<50% correct alv stops			
		- =/<10th percentile CDI	11	4	5
		- Total	13	5	8
	No need for intervention		*0*	*8*	*5*
CONT group	Need for intervention	- No alveolar stops	9	7	10
		<50% correct alv stops			
		- =/<10th percentile CDI	11	7	6
		- Total	13	11	*11*
	No need for intervention		*0*	*2*	*2*

In the present study, the speech criterion for continued need for intervention was non-emerging alveolar stops (less than 50% correctly produced alveolar stops) as opposed to absence of alveolar stops at pre-test and four months after intervention (Jørgensen et al., in preparation). Across groups, nineteen/26 toddlers produced less than 50% alveolar stops correctly in the present study whereas only 11/26 toddlers did not produce alveolar stops in Jørgensen et al. (in preparation). As very few toddlers with CP had age-typical speech accuracy scores, it seems that the speech criterion in the present study matched speech performance better than the original criterion.

Clinical implications

Results from this study showed that parent-implemented FS did not have long-term effect on speech accuracy, receptive language, or need for further intervention in toddlers with CP and delayed speech and/or language development. No study to date has documented long-term effects of early intervention in toddlers with CP. Albeit, these results suggest that early intervention in toddlers with CP may simply not be worthwhile, there are many possible explanations why long-term effects were not found in this study. In particular, the CONT group in the present study seemed to catch up on productive vocabulary for age. Short-term effect on productive vocabulary is the only effect reported across all previous intervention studies in toddlers with CP including control groups (Ha, 2015; Kaiser et al., 2017; Jørgensen et al., in preparation). In light of the present result, it seems worth considering whether productive vocabulary is an appropriate intervention goal for toddlers with CP and small vocabularies. Speech recasting may be more efficient in children with larger vocabularies as opportunities for recasting are greater or at least easier identifiable for clinicians or parents. Hence, more advanced speech-language development may be a relevant prognostic factor for intervention effect on speech accuracy in children with CP, as also pointed out by Kaiser et al. (2017). Although optimal intervention intensity is not easily determined (Warren, Fey, & Yoder, 2007), expanding total intervention duration and perhaps dose frequency seems relevant for obtaining long-term intervention effects in toddlers with CP as these toddlers may need more production practice than toddlers with speech-language delay without CP. For the same reason, it should also be considered whether toddlers with CP may benefit more from intervention that require child responses, such as EMT, in terms of effect on speech accuracy.

Limitations

The present study involved relatively small groups of toddlers with CP. Small group sizes may influence results, particularly in studies involving heterogeneous populations such as toddlers with CP. Further, this study included toddlers with mixed cleft types. In general, speech seems to be more affected in more extensive clefts (Hardin-Jones, Chapman, & Schulte, 2003; Lohmander-Agerskov, Söderpalm, Friede, Persson, & Lilja, 1994; Persson, Elander, Lohmander-Agerskov, & Söderpalm, 2002). The issues of both mixed cleft types and heterogeneity were to some extent addressed by introducing speech-language criteria. Hearing tests were not available and thus it not known if hearing status differed between groups. However, presence of OME and early PET insertion were highly similar in the two groups.

The present study presents the long-term results of a previous study (Jørgensen et al., in preparation). However, it was not possible to determine whether the statistically significant effects that were found in spontaneous speech-language samples four months after pre-test (gain in lexical age for observed productive vocabulary and gain in true consonant types in spontaneous word productions) were still present six months after termination of intervention because the present study only included information from the CDI and transcriptions of words from a naming test.

It is beyond questioning that long-term results of intervention are desirable; notwithstanding, they are also very hard to interpret. Children with need for intervention will, in all likelihood, be enrolled in other interventions, introducing a confounding variable that is difficult to control for. Six toddlers in this study's CONT group attended local speech therapy services where the local SLP met occasionally with parents or preschool teachers. Although this may in theory have influenced outcomes in the CONT group, studies of adult learning (Trivette, Dunst, Hamby, & O'Herin, 2009) and the reported inefficiency of local speech therapy services in toddlers with CP (Hardin-Jonas & Chapman, 2008) would suggest that such services are not ultimately effective in changing child behaviour.

Conclusion

Parent-implemented FS did not have long-term effect on speech accuracy, reported productive vocabulary, receptive language, or need for further intervention. Very few toddlers with CP reached the speech accuracy levels of their peers without CP. Several factors may explain these findings. However, careful consideration of intervention method, dose frequency and total intervention duration, and intervention targets is recommended before provision of early intervention to toddlers with CP and delayed speech and/or language development. Future studies of early intervention in toddlers with CP should include longitudinal follow-up.

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Appendix 1. WSSA analyses of the word 'bamse' (*Eng. 'teddy bear'*) for three different participants.

Orthography	'	b	a	m	s	e	'
Target	/	b̥	ɑ	m	s	ə	/
Actual1	[	b̥	ɑ		s	ə	]

Global structural agreement: One orphan slot; (4/5) = 0.8.

Featural agreement: No disagreement on any of the two consonants (vowels counted as correct); 4/4=1.0.

WSSA: 0.8 x 1.00 = 0.8.

Orthography	'	b	a	m	s	e	'
Target	/	b̥	ɑ	m	s	ə	/
Actual2	[	h	ɑ	f	s	ə	]

Global structural agreement: No orphan slots; (5/5) = 1.0.

Featural agreement: /b̥/ → [h]: big manner deduction of .25, major place deduction of .333, no voicing deduction; featural agreement = 1 - 0.25 - 0.333 = 0.417.

/ɑ/ → [ɑ]: No deduction; featural agreement = 1.0.

/m/ → [f]: major manner deduction of .25, teeny place deduction of .0833, major voicing deduction af .333; featural agreement = 1 - 0.25 - 0.833 – 0.333 = 0.3337.

/s/ → [s]: No deduction; featural agreement = 1.0.

/ə / → [ə]: No deduction; featural agreement = 1.0.

Mean featural agreement: (0.417 + 1.0 + 0.334 + 1.0 + 1.0) / 5 = 0.75.

WSSA: 1.0 x 0.75 = 0.75.

Orthography	'	b	a	m	s	e	'
Target	/	b̥	ɑ	m	s	ə	/
Actual3	[	m	əː				]

Global structural agreement: Three orphan slots; (2/5) = 0.4.

Featural agreement: /b̥/ → [m]: major manner deduction of .333, no place deduction, major voicing deduction of .333; featural agreement = 1 - 0.333 - .333 = 0.333.

/ɑ/ → [əː]: No deduction (vowels counted as correct); featural agreement = 1.0.

Mean featural agreement: (0.333 + 1.0) / 2 = 0.667.

WSSA: 0.4 x 0.667 = 0.267.

Appendix 2. List of test items in TEFF.

TEFF-1				TEFF-2 (changes from TEFF-1)			
WI target	Word	Translation	IPA	WF target	Word	Translation	IPA
/p-/	pude *pige penge	pillow girl money	[ˈpʰuːð̞ə] [ˈpʰiːi] [ˈpʰɛŋə]				
/t-/	*tog *tom *tiger	train empty/all gone tiger	[ˈtˢɔw] [ˈtˢʌmˀ] [ˈtˢiːɑ]				
/k-/	*ko *kage *kop	cow cake cup	[ˈkʰoˑˀ] [ˈkʰæːæ] [ˈkʰʌb̥]				
/b-/	*bil *bamse *bog	car teddy bear book	[ˈb̥iˑˀl] [ˈb̥ɑmsə] [ˈb̥ɔwˀ]	/-b/	*rap *kop	quack cup	[ˈʁɑb̥] [ˈkʰʌb̥]
/d-/	dyne *dør *juice[1]	duvet door juice	[ˈd̥yːnə] [ˈd̥œʌ̯ˀ] [ˈd̥juːs]	/-d/	*sut *kat	pacifier cat	[ˈsud̥] [ˈkʰad̥]
/g-/	gynge *gul *gave	swing yellow present	[ˈg̥øŋə] [ˈg̥uˑˀl] [ˈg̥æːwə]	/-g/	*tak *æg	thank you egg	[ˈtˢɑg̥] [ˈɛˑˀg̥]
/f-/	*fugl *far *får	bird father sheep	[ˈfuˑˀl] [ˈfɑː] [ˈfɒˑˀ]	/-f/	*giraf	giraffe	[g̥iˈʁɑf]
/s-/	*sove *sut cykel	sleep pacifier bicycle	[ˈsɒːwə] [ˈsud̥] [ˈsyg̥əl]	/-s/	*is *bus	ice cream bus	[ˈiˑˀs] [ˈb̥us]
/ʋ-/	*vand vindue *vante	water window mitten	[ˈʋanˀ] [ˈʋend̥u] [ˈʋand̥ə]				
/l-/	*lys *løve lastbil	candle/light lion truck	[ˈlyˑˀs] [ˈløːwə] [ˈlas(d̥)ˌb̥iˑˀl]	/-l/	*bil *sol	car sun	[ˈb̥iˑˀl] [ˈsoˑˀl]
				/-ð/	*pude *rød	pillow red	
/j-/	*ja jakke jordbær	yes jacket strawberry	[ˈja] [ˈjɑg̥ə] [ˈjoʌ̯ˌb̥æʌ̯]				
/ʁ-/	*rap *rutsje *rød	quack slide red	[ˈʁɑb̥] [ˈʁo(d̥)ɕə] [ˈʁœð̞ˀ]				
/m-/	*mælk *mor *mus	milk mother mouse	[ˈmɛlˀg̥] [ˈmoːɑ] [ˈmuˑˀs]	/-m/	*bamse arm	teddy bear arm	[ˈb̥ɑmsə] [ˈɑˑˀm]
/n-/	*nej næse	no nose	[ˈnɑjˀ] [ˈnɛːsə]	/-n/	*and *hund	duck dog	[ˈanˀ] [ˈhunˀ]

	navle	belly button	[ˈnɑwlə]				
				/-ŋ/	gynge *seng	swing bed	[ˈg̊øŋə] [ˈsɛŋˀ]
/h-/	*hund *hue *hus	dog hat house	[ˈhunˀ] [ˈhuːu] [ˈhuˑˀs]				

*Words used for WSSA analysis. [1]Young Danish children will typically pronounce the first consonant in true clusters.

Note: Danish syllable final approximants, except for /ð/, are semivowels and not considered as target consonants in this study.

Appendix 3. PCC-A, WSSA, CDI, and NRDLS standard scores for all participants.

INT GROUP					CONT GROUP				
Child	PCC-A	WSSA	CDI	NRDLS SS	Child	PCC-A	WSSA	CDI	NRDLS SS
1	0.20	0.60	78	65	1	0.52	0.82	265	68
2	0.14	0.63	120	104	2	0.64	0.88	353	93
3	0.62	0.87	426	91	3	0.55	0.86	233	114
4	0.47	0.79	298	99	4	0.64	0.89	249	78
5	0.87	0.95	488	104	5	0.51	0.80	561	93
6	0.60	0.89	335	101	6	0.69	0.88	221	74
7	0.40	0.80	576	88	7	0.39	0.74	256	73
8	0.65	0.86	128	91	8	0.43	0.84	243	79
9	0.45	0.83	258	74	9	0.31	0.73	93	102
					10	0.06	0.55	5	70
11	0.20	0.70	296	93	11	0.41	0.77	259	81
12	0.59	0.87	246	84					
13	0.29	0.70	199	92	13	0.47	0.78	436	77
14	0.33	0.74	251	110	14	0.24	0.68	125	86

DISCUSSION

The studies in this dissertation addressed the following main questions:

1. Can early delays in vocabulary and/or speech development in toddlers with CP be positively affected by parent-implemented focused stimulation?

2. Can need for intervention in toddlers with CP be assessed in a reliable and valid, yet time-efficient, way?

Studies 2 and 3 addressed the first question. Study 2 focused on the short-term intervention effects of productive vocabulary and consonant inventory while study 3 was concerned with long-term effects on speech accuracy, reported productive vocabulary, and receptive language. Study 1 aimed to address the second question by evaluating a screening procedure for identifying intervention need. The main results of the three studies are summarised below. Overall, these studies provide new insights into both the short-term and long-term efficiency of parent-implemented focused stimulation in toddlers with CP and speech and/or language delay as well as a framework for designing intervention studies in this clinical group.

Study 1 described the development and evaluation of a clinically useful procedure for screening of speech-language development in toddlers with CP. The screening procedure combined information on parent-reported productive vocabulary from the CDI with information on consonant inventory from real-time listening (RTL) to determine need for intervention. CDI and RTL were selected because they could provide information on measures of productive vocabulary and consonant inventory which correlated with later speech-language development in longitudinal studies of toddlers with CP. In addition, CDI and RTL were time-efficient assessment methods compared to standard assessment procedures of speech and language development in toddlers. Although the predictive measures of productive vocabulary and consonant inventory had to be amended to fit CDI and RTL assessment, reliability and external validity of the procedure was demonstrated in 22 toddlers with CP. Good-excellent inter-rater agreement between three trained raters confirmed the hypothesis that RTL was a reliable method for screening consonant inventory in toddlers with CP. The hypothesised substantial agreement between the screening procedure and a clinical reference standard was also confirmed as two experienced SLPs agreed almost perfectly with the screening procedure in determining who needed intervention, thus indicating high external validity of the screening procedure.

Study 2 investigated the short-term effects of parent-implemented focused stimulation. Both vocabulary and consonant inventory were directly targeted. Parents of 14 toddlers with CP and delayed speech and/or language development were taught language support strategies. These

14 toddlers were compared to a pairwise matched control group of 14 toddlers with CP. Intervention effect was found for lexical age, a measure of observed productive vocabulary development. Hence, the hypothesis that intervention would affect rate of vocabulary acquisition was confirmed. Intervention effect was also found for true consonant types in words. Interestingly, this effect on consonant inventory could largely be explained by the intervention group's increased ability to produce words with true consonants already within their consonant inventory. Thus, the hypothesis that consonant inventory would expand as a function of vocabulary expansion was only partly confirmed as use of consonant inventory did seem to expand whereas new consonants were not added to inventory in all vocalisations beyond the effect of general development. This result indicates that it may be beneficial to select target words based on within-inventory consonants as adding target words with outside-inventory consonants did not seem to facilitate use of these consonants. The intervention group was significantly more likely to not need intervention at post-test compared to the control group. Need for intervention was based on whether participants still met the pre-test inclusion criteria at post-test. As these criteria were based on predictive measures of later speech-language difficulties, this result may have positive implications for later speech-language development. A parent questionnaire revealed that parents in general found that intervention had positively affected their child's speech-language development, and that parents were satisfied with the intervention program. In addition, most outcome measures increased whether or not the participants received intervention which confirms the effect of general development and highlights the importance of including matched control groups in intervention studies in this age group.

Study 3 reported the long-term results of parent-implemented focused stimulation six months after termination of intervention. It was hypothesised that short-term effects of home-based parent-implemented intervention would be maintained because parents were taught to use language support strategies in naturally occurring interactions with their child. This hypothesis was not confirmed as no intervention effect was found for any of the outcome measures: speech accuracy, reported productive vocabulary, receptive language, or need for intervention, and as very few toddlers with CP obtained speech accuracy scores within 2 standard deviations of the mean for TD toddlers. It was to some degree expected that intervention would not affect speech accuracy as the hypothesis of Study 2 that toddlers in the intervention group would expand their consonant inventories to a larger degree than toddlers in the control group was only partly confirmed. However, since Study 2 showed an intervention effect on true consonant types and a large effect size for oral stop tokens in words, an intervention effect on PCC-A for true consonants or oral stops in Study 3 could be hypothesised, but was also not confirmed. Although these results suggest that early intervention in toddlers with CP may simply not be worthwhile, there are many possible explanations why long-term effects were not found in study 3. These include choice of outcome measures and speech- language samples, language level of participants, intervention method and facilitator, and cumulative intervention intensity. Nonetheless, the results of Study 3 warrants careful consideration before provision of early intervention to toddlers with CP and delayed speech and/or language development.

STRENGTHS AND LIMITATIONS OF THIS PROJECT

One of the major strengths of this project is that participants were selected from a complete cohort of toddlers with CP based on predetermined general inclusion criteria and speech-language inclusion criteria. This minimised the risk of sampling bias which is often seen in studies of individuals with CP (Shaw et al., 2016). However, a different kind of sampling bias was introduced because participants were not randomly allocated into the intervention group and the control group. Non-random group allocation was chosen for practical reasons, but random group assignment in this project would also have raised ethical concerns as providing early intervention to some toddlers but not others, when they were seen in the same cleft clinic, would have been difficult to justify. The non-random intervention design probably led to more parents in the Aarhus centre declining participation, but may also explain why no families in this project were lost to attrition.

Heterogeneity and general development are two powerful factors affecting group comparison of speech-language development of toddlers with CP. Thus, participants were pairwise matched on reported vocabulary size and reported vocabulary size for age in an attempt to strengthen experimental control and reduce the risk of bias introduced by non-random group allocation. At group level, participants were matched for age, age at palatal surgery, cleft type, and SES, but not time from palatal surgery to pre-test. A concern regarding participant selection is that two toddlers with cleft lip and unoperated submucous cleft palate were part of the control group in Studies 2 and 3. These participants were included because they matched the speech-language inclusion criteria and thus reflected the toddlers with operated CP. On one hand, they may have had VPD and needed palatal surgery, but both these toddlers produced oral stops which makes VPD less likely. On the other hand, their speech-language delay may have had a different underlying aetiology than CP. However, this may be true for all participants in Studies 2 and 3. Ideally, more participants should have been included to allow for even more careful matching. Nonetheless, the matching in the present study is probably as close to optimal as possible in this clinical population because participants were selected from a complete cohort of 103 toddlers with CP.

To observe the true effect of general development, the control group should have received no treatment at all, but again, this would have been unethical. Hence, it is possible that a true non-treatment control group would have exhibited a slower rate of development than the control group in Study 3. Nonetheless, theories on adult learning strategies would indicate that counselling in itself is not adequate to change parents' use of language support strategies (Trivette, Dunst, Hamby, & O'Herin, 2009), and both clinical experience from Denmark as well as the results of the retrospective studies by Hardin-Jones and Chapman (2008) and Raud Westberg et al. (2017) suggest that early speech therapy services are not ultimately effective in changing child behaviour toddlers with CP.

A particular strength of Study 2 is that fidelity measures were reported to account for the triadic nature of parent-implemented intervention. Thus, it was documented that parents were able to use language support strategies taught in intervention with their child. However, parents' continued use of strategies after intervention was not reported. Although the aim of Study 3 was to investigate the long-term effects of an intervention program that lasted three months, it was also assumed that parents would continue to use the language support strategies that they had learned during intervention. If parents did not continue to use strategies, that may be responsible for the null-findings in Study 3.

The strength of Study 1 is that identification of need for intervention was based on predictive measures of later speech-language difficulties, and that evaluation of need for intervention by expert SLPs served as the clinical reference standard for this screening procedure. It is a limitation that the predictive measures had to be amended to fit into the time-efficient assessment of parent report and RTL because it is unknown whether the amended measures were still predictive. Further to this point, long-term sensitivity and specificity of need for intervention were not reported. However, Study 3 revealed that all but two out of fourteen toddlers in the control group still met the slightly altered speech-inclusion criteria ten months after pre-test. This may indicate that the screening procedure has high long-term sensitivity although this result was not validated against the opinion of expert SLPs. Long-term specificity is not known because consonant inventories of toddlers who did not need intervention at pre-test were not transcribed. Thus it is not known whether these toddlers still did not need intervention at a later time point. Hopefully, future analyses of the data from these toddlers will answer this question and help validate the specificity of the screening procedure.

Intervention effects are dependent on the selection, assessment, and evaluation of outcome measures. Studies 2 and 3 use well-defined outcome measures and report inter and intra-rater reliability. The rigorous methodology enables future replication of studies while comparison with earlier studies is difficult because earlier studies either applied different outcome measures or did not provide accurate descriptions of these. However, in toddlers, it is difficult to construct outcome measures that adequately reflect development, in particular when comparing groups. The lexical age measure was constructed to account for different developmental rates in the early vocabulary and was used in Study 2. However, because many children had expanded their vocabulary considerably ten months after pre-test, the lexical age measure could not be applied in Study 3.

Ideally, assessment and evaluation of outcomes should be performed by a person blinded to both study purpose and group allocation. In this project, the author did all assessments of the participants in Copenhagen and another SLP did all assessments in Aarhus. Thus, only the second SLP was blinded to study purpose and group allocation. It is most likely that lack of blinding was not a major source of bias during the spontaneous parent-child interaction assessments where the SLP did not interfere, but both the receptive language test and the naming test may have been confounded. All SLPs and students involved in evaluations of outcomes, except the author, were blinded to both study purpose and group allocation in Study 1, and as a minimum blinded to group allocation in Studies 2 and 3. All evaluations, including those rated by the author, were re-evaluated by at least one other rater to ensure inter-rater reliability, but also to minimise the risk of bias introduced by the incomplete blinding procedure during evaluation.

Studies 2 and 3 reported the short-term and long-term effects of intervention, respectively. Different outcome measures were reported in the two studies. Study 2 reported the results from spontaneous speech assessment whereas Study 3 reported the results from a naming test. Hence, it is not possible to determine whether the intervention effects on gain in true consonant types in words and lexical age of observed productive vocabulary were maintained. It was hypothesised that these short-term effects may have affected the measures of speech accuracy and reported vocabulary included in Study 3, but this was not confirmed. A long-term measure of observed vocabulary would have been informative and highly relevant.

In this project, statistical significance was reported along with measures of effect sizes. Significant results and large effect sizes were interpreted both in the context of previously reported intervention effects and in relation to clinical relevance. In addition, confidence intervals were also reported. Confidence intervals were generally wide in all the included studies. As the range of confidence intervals depend on sample size, it is likely that wide confidence intervals reflect the relatively small group sizes. Thus, the validity of results with wide confidence intervals in this project would clearly benefit from replication of the studies.

Additional investigations

When Study 1 was designed, all participants had not been assessed at pre-test. Later, however, pre-test data from all participants became available. As the screening procedure managed so well to identify the toddlers with CP who needed intervention, these data provided an opportunity to look closer at factors other than speech and language that may be characteristic of toddlers with CP who need or do not need early intervention. These factors may potentially strengthen the prediction value of the screening procedure.

The first measure that may be relevant is age. Speech-language development will be evident as toddlers grow older, and both measures of vocabulary and consonant inventory have been found to increase in toddlers with CP, regardless of developmental level (Chapman, 2004; O'Gara & Logemann, 1988; Willadsen, 2012). Thus, it is possible that the screening procedure will be more likely to find that younger participants need intervention than older participants. If this is the case, the true need for intervention may be overestimated as participants in the "need for intervention" category may later evidence development that deems intervention unnecessary. In this project however, participants who needed intervention were actually slightly older (mean age: 19.18 months) than those who did not need intervention (mean age: 18.95 months).

Gender may also be a highly relevant factor. One of the best predictors of being a late talker is being a boy (see Rescorla, 2011), and at group level, early productive vocabulary development is slower in boys than in girls (Bleses et al., 2008). Studies of speech sound acquisition in different languages report conflicting results with some finding that girls perform better than boys (Dodd, Holm, Hua, & Crosbie, 2003; Phoon, Abdullah, Lee, & Murugaiah, 2014; Smit, Hand, Freilinger, Bernthal, & Bird, 1990) and some finding no difference (Fox, 2006; Maphalala, Pascoe, & Smouse, 2014). In a recent study of speech sound acquisition in Danish children aged 2;6 to 4;11 years, no influence of gender was reported (Clausen & Fox-Boyer, 2017). In children with CP, Willadsen et al. (2017) recently reported that boys with unilateral CLP perform poorer than girls on measures of speech accuracy at three years, and that boys' speech development seems to be more vulnerable to an unoperated hard palate than girls'. In the present project, boys were more likely to need intervention than girls as 25 boys needed intervention and five did not, while eight girls needed intervention and 17 did not, indicating that more boys than girls with CP need intervention.

Participants in the present project had mixed cleft types. Although evidence is scarce, there may be a difference in speech-language performance between cleft types. It seems that the less extensive the cleft, the lesser influence on speech. Hence, children with complete bilateral CLP are likely to have poorer speech than children with an isolated cleft of the soft palate (Hardin-

Jones, Chapman, & Schulte, 2003; Lohmander-Agerskov et al., 1994; Persson, Elander, Lohmander-Agerskov, & Söderpalm, 2002). In this project, 26 participants with CLP needed intervention and ten did not, whereas seven participants with CPO needed intervention and 12 did not. This may indicate that toddlers with CLP are more likely to need intervention than toddlers with CP.

Age at palatal surgery is likely to reflect speech outcome, however, since most participants in the present project had surgery at fairly similar ages, this factor is not expected to influence intervention need in this project. Participants who needed intervention were older when they had surgery (mean age: 13.42 months) than those who did not need intervention (mean age: 12.25). Four participants had cleft lip and submucous cleft palate and had not received palatal surgery. These four participants were distributed evenly in the two groups. A related factor is for how long participants have had an operated palate, i.e. time passed from surgery to pre-test. This may be a relevant factor since it may take up to eight weeks before catch-up to pre-surgical speech level, and before further development can occur (Grunwell & Russell, 1987). However, all participants had had a complete palate for at least eight weeks, indicating that time passed from surgery to pre-test would not affect need for intervention in this project. Participants who needed intervention had had an operated palate for a shorter period of time (mean time: 5.74 months) than those who did not need intervention (mean time: 6.65 months).

Some studies indicate that SES may influence vocabulary size (e.g. Fernald, Marchman, & Weisleder, 1998), but SES generally explains very little variance in language performance in toddlers (Rescorla, 2011). Thus, SES is not expected to influence need for intervention in this project. Both participants who needed intervention and those who did not had a median SES of 3 (lower middle class).

Table 7 shows Pearson's r and Spearman's ρ correlations between need for intervention and the participant characteristics age, gender, cleft type, age at surgery, time from palatal surgery to pre-test, and SES.

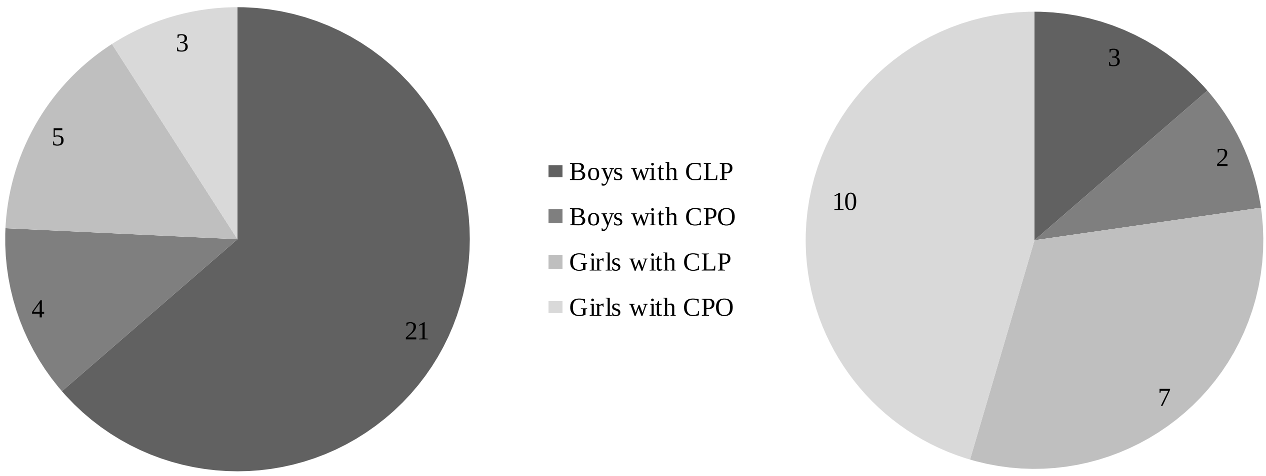
Table 7. Correlations between need for intervention and different participant characteristics.

	Age	Gender	Cleft type	Age at surgery	Time from surgery to pre-test	SES
Need for intervention	-.04	.55**	.34*	-.27	.15	.08

**$p < 0.001$, * $p < 0.05$.

Statistically significant correlations were found for gender (moderate size) and cleft type (small size), confirming that boys are more likely to need intervention than girls, and that toddlers with CLP are more like to need intervention than toddlers with CPO. Because boys are more likely to have CLP than girls whereas girls are more likely to have CPO, these correlations may be explained by interaction between gender and cleft type. The distribution of gender and cleft type in the two groups is illustrated in figure 5. However, a three-way loglinear analysis indicated that the highest-order interaction (gender*cleft type*need for intervention) was not statistically significant, $X^2(1)=0.079$, $p=.778$. This means that boys are more likely to need intervention than girls irrespective of cleft type, and that toddlers with CLP are more likely to need intervention than toddlers with CPO irrespective of gender.

Figure 5. Distribution of gender and cleft type in toddlers who needed intervention (left) and toddlers who did not need intervention (right).



Although no significant correlation was found for age, a possible interaction between age and gender was investigated. It was speculated that younger boys were more likely to need intervention based on the screening procedure than older boys and girls because of slower general development in boys (Bornstein, Hahn, & Haynes, 2004; Fenson et al., 2007). Logistic regression analysis to predict need for intervention using gender and a possible interaction of gender*age at pre-test as predictors could not be performed due to small cell counts. Thus, a possible influence of interaction of gender and age at pre-test on need for intervention was explored by looking at the mean ages of boys and girls respectively in each group. While there was clearly no difference for girls (mean age: need for intervention: 19.62 months; no need for intervention: 19.65 months), there was a larger difference for boys who needed intervention (mean age: 19.04 months) and boys who did not (mean age: 16.60 months). Interestingly, boys who did not need intervention were younger than the boys who needed intervention and therefore, the hypothesis of slower general development was not confirmed. It should be noted, however, that only five boys did not need intervention and thus these results should be interpreted with caution.

FUTURE DIRECTIONS AND CLINICAL IMPLICATIONS

If early intervention can prevent later speech-language difficulties, and if young children who will have persistent speech-language difficulties can be reliably identified, then naturally, the cost and burden of care of early speech-language intervention would be justified. In children without CP, the challenge is reliable prediction of later speech-language difficulties whereas the effect of early intervention is well-documented, although long-term results are less commonly reported. In children with CP, however, it may be the other way round. The results of the studies reported here suggest that need for intervention in toddlers with CP can be identified, but that early speech-language intervention may not have the same effect as in other clinical groups, at least in a long-term perspective.

The paramount question is of course whether toddlers with CP and speech-language delay should be enrolled in early intervention. The results of this project clearly imply that early intervention is not a quick-fix for toddlers with CP. Yet, many questions remain unanswered.

First of all, the observed short-term effects on vocabulary and consonant inventory in words document the potential benefit of parent-implemented FS, but also raise the question how these effects can be maintained or even expanded. Possible answers may be a prolonged follow-up period or introducing a different kind of intervention. Another question is whether including only target words with consonants within inventory may facilitate even greater vocabulary expansion, and whether this in turn will lead to long-term effect on vocabulary development. Further, it is an open question whether it is necessary to target vocabulary separately. Many toddlers in the control group had outgrown vocabulary delay ten months after pre-test whereas speech delay was still prevalent.

Comparison with earlier studies of intervention in toddlers with CP is not straightforward. Even so, the study by Kaiser et al. (2017) raises the question whether clinician-implemented EMT twice/week for six months have greater impact on speech accuracy and receptive language in toddlers with CP and more advanced language development compared to the present project. Unfortunately, it is not possible to separate the effects of intervention method, facilitator, total intervention duration and dose frequency, and language development.

In addition to language development level, other participant characteristics, e.g. severity of speech delay or combined receptive and productive language delay, may also be factors that influence prediction of intervention outcome. More early intervention studies involving toddlers with CP are needed before it can be determined what works and what works better for whom.

This project presents a framework for designing future early intervention studies in this clinical group. The centralised Danish model for treatment of CP enabled inclusion of a complete cohort of toddlers with CP in this project, but this model is not common around the world. Hence, researchers face different challenges when designing intervention studies in this clinical population. Still, speech-language inclusion criteria as proposed in this project and careful matching of participants may greatly enhance the quality of future early intervention studies.

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