

A multimethod study of the impact of stuttering on children

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Abstract

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List of papers

Paper I: Guttormsen, L. S., Kefalianos, E., & Næss, K. A. B. (2015). Communication attitudes in children who stutter: A meta-analytic review. *Journal of Fluency Disorders*, 46, 1–14.

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Note. These papers follow the extended abstract.

1. Introduction

1.1 Background

Playing soccer; meeting a new friend; talking to mum before going to bed; telling the kindergarten teacher a story; quarreling with siblings. Stuttering influences the most essential aspect of human interaction: communication. For those affected, stuttering can be a far-reaching disorder. As described by many authors within the field (e.g., Guitar, 2014; Shapiro, 2011; Ward, 2008; Yaruss & Quesal, 2006), stuttering is a complex phenomenon, characterized not only by moments of stuttering but by the various consequences of these speech disruptions.

In recent decades, a number of studies have investigated the consequences of stuttering for the youngest affected population: children aged two to six years (referred to hereafter as *young children*). While investigations of impact have depended mainly on parental reports (e.g., Langevin, Packman & Onslow, 2010; Millard & Davis, 2016), studies targeting communication attitudes have commonly used self-reporting from the age of three (e.g., Brce & Vanryckeghem, 2017; Clark et al., 2012; Vanryckeghem, De Niels, & Vanrobaeys, 2015).

Langevin et al., (2010) suggested that treatment should be initiated if children are affected by the stuttering. This because several treatment programs have shown to be effective in treating stuttering in young children (see for instance de Sonnevile-Koedoot, Stolk, Rietveld, & Franken, 2015; O'Brian et al., 2013), and thereby reducing or eliminating the negative consequences of the disorder. If impact is to be useful as an indicator of treatment need, valid and reliable assessment of all relevant aspects of impact in young children is essential.

1.2 Aim and outline of the thesis

The overarching aim of this thesis is to investigate how children are affected by stuttering. Because of the apparent inconsistencies and variations in research findings, instruments and methods, the aim was to locate and summarize the body of research on communication attitudes and to examine caregivers' perceptions of overall impact and the reliability of these

reports. To that end, the project has two general objectives: 1) to investigate how stuttering affects young children's lives, and 2) to investigate the reliability of caregiver report of impact of stuttering.

The thesis comprises an extended abstract and three articles. As shown in Figure 1, each paper contributes differently to the overarching aim by application of different methods and informants.

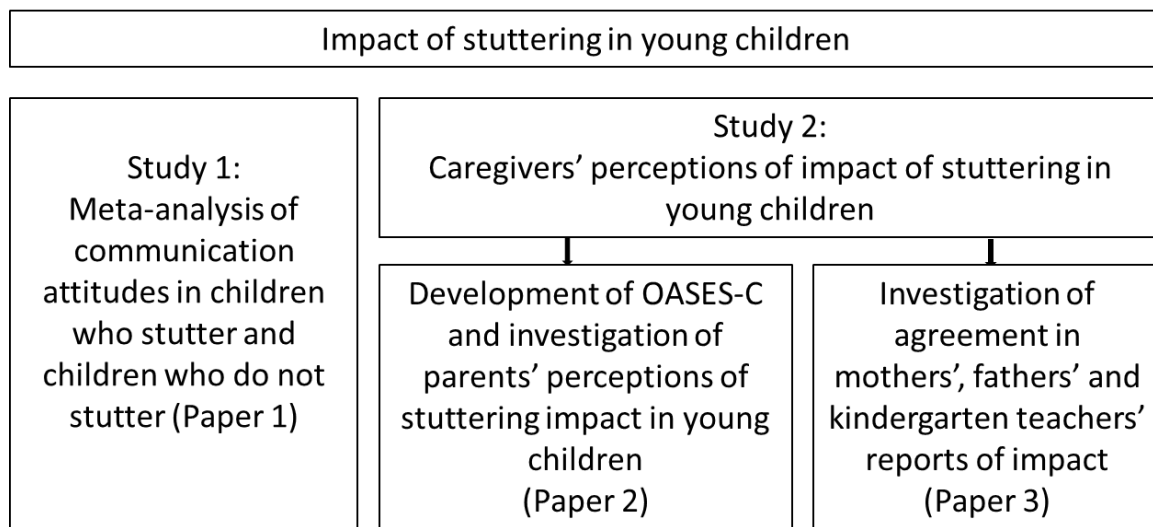


Figure 1. Overview of the studies and associated papers in the thesis.

1.3 Construct clarification

The construct of *impact*, referring to the different effects of stuttering, is variously operationalized and defined across different studies. For present purposes, Yaruss and Quesal's (2010) definition is applied (see section 3.1 for review of the literature). On this definition, impact includes children's emotional, behavioral and cognitive reactions to stuttering; difficulties when communicating; and reductions in quality of life (QoL). Importantly, social interaction, self-confidence, mood and participation are included as components of quality of life. In the relevant literature, both cognitive reactions to stuttering (see Yaruss and Quesal, 2016), and communication attitudes (see for instance Vanryckeghem) reflect children's thoughts regarding their speech (as further discussed in sections 3.2 and 5.2.2). Though operationalized differently, both constructs indicate negative speech-related thoughts and attitudes—the cognitive aspect of stuttering impact. In this thesis, the construct that corresponds to the operationalization in the different studies will be

used when the studies are reviewed (e.g., reporting on studies that have applied the KiddyCAT, the construct communication attitudes will be used).

As kindergarten and preschool ages differ by country, the term *young children* is applied here to children aged two to six years. The term *kindergarten teachers* refers here to pedagogically educated staff working with young children in childcare centers.

2. Stuttering

2.1 What is stuttering?

At first, stuttering appears to be complex and mysterious (Guitar, 2014). The uniqueness of stuttering as a disorder relates to its sudden emergence in speech of normal fluency (Packman & Attanasio, 2017) and, for many children, its equally sudden resolution. A further intriguing feature of the disorder is its natural variability; present in periods and absent in others; appearing in specific sounds and words but not others; and disappearing completely under fluency-inducing conditions (e.g., rhythmic speech). The literature concerning the nature of stuttering will be reviewed in this chapter, but firstly a definition of the disorder under study is provided.

2.1.1 Definition of stuttering

A common understanding of stuttering is that it involves disruptions in speech, even though the speaker knows what they want to say. For communication partners, stuttering is easily detected, but it remains challenging to define the disorder, and it has long been recognized that there is no adequate definition. Packman and Attanasio (2017) noted that beyond the usual challenges of defining disorders, certain features of stuttering make it especially difficult to achieve consensus on an adequate definition of stuttering. These features relate to the frequency and variability of stuttering behavior and the distinction between stuttering behavior and normal speech disfluency. The lack of an adequate definition of stuttering may either be looked upon as academic or even trivial, or as an obstacle for conducting research of stuttering (Packman & Attanasio, 2017) (see 5.1.1 for a discussion of operationalization of stuttering). According to Onslow (December, 2017) among the many attempts to define stuttering—in terms of observer perceptions, speaker experiences or dictionary definitions of symptoms—the World Health Organization definition from 1972 is most commonly applied: “Disorders of the rhythm of speech, in which the individual knows precisely what he wishes to say, but at the time is unable to say it because of an involuntary repetition, prolongation or cessation of a sound” (p. 202).

2.2 The aetiology of stuttering

Many researchers would agree with Chang et al.'s (2017) conclusion that stuttering is a complex neurodevelopmental disorder. The challenge, however, is to specify the factors involved in the development of stuttering and their contribution to the disorder. Because of the disorder's inherent complexity, theories differ in what they seek to explain; while some target the conditions present at the onset of stuttering, others focus specifically on factors that cause the individual moments of stuttering (Bloodstein, 1995). However, a common feature of many theories is that they are multifactorial and focus on the interplay between the vulnerable speech motor system and environmental, emotional and linguistic factors. Examples include the dual diathesis stress model (Walden et al., 2012); the demands and capacities model (Starkweather & Gottwald, 1990); and the integrated perspective on stuttering (Guitar, 2014). Recently, the Multifactorial dynamic pathways theory was proposed by Smith and Weber (2017). The MDP theory explains critical features of the stuttering disorder such as onset and development, natural recovery and the role of genetics in development of stuttering. This theory is supported by extensive research and will therefore serve as a theoretical framework for the following review on the literature of the aetiology of stuttering.

2.2.1 Multifactorial dynamic pathways theory

In line with the DSM-5 (APA, 2013), Smith and Weber (2017) defined stuttering as a neurodevelopmental disorder, in which “the primary symptoms of stuttering are dysfluencies, involuntary disruptions in the normal flow of speech” (p. 2485). The premise of multifactorial dynamic pathways (MDP) theory is that stuttering is caused by several different factors. Importantly, as the combination of these factors can differ across individuals and across time, children's pathway into stuttering and the development of stuttering is dynamic.

More specifically, MDP theory asserts that the mechanism that produces stuttering is a central nervous system failure to generate the patterns of motor commands necessary for fluent speech production. Smith and Weber also emphasized that the occurrence, developmental pathway (recovery versus persistency) and severity of stuttering is strongly conditioned by linguistic and emotional factors.

2.2.2 Genetics

In recent decades, documented evidence of a genetic predisposition to stuttering (Packman & Attanasio, 2017) includes higher concordance rates for monozygotic twins (Dworzynski, Remington, Rijdsdijk, Howell, & Plomin, 2007) and higher frequency of stuttering relatives among kindergarten children who stutter (Ambrose, Yairi, & Cox, 1993). Felsenfeld et al. (2000) attributed approximately 70% of the variance in liability to stuttering to genetic factors. This would suggest that children can inherit a neural system that is more susceptible to fluency breakdown. However, as Dworzynski, et al. (2007) showed that the majority of monozygotic twin pairs were discordant rather than concordant, this necessary genetic predisposition alone is not sufficient for developing stuttering. There are several gaps in the literature on the genetic contribution to the onset of stuttering and Yairi and Ambrose (2013) related this lack of understanding to the polygenic nature of stuttering. Smith and Weber (2017) highlighted the role of epigenesis; that is, the timing and intensity of gene expression over development.

2.2.3 Neurological underpinnings of stuttering

According to Smith and Weber (2017), the emergence of stuttering is not determined at birth, as the structure of the brain is always a result of the interaction between genes, epigenetic processes and individual experiences. Many studies have investigated the neurological signature of stuttering, often focusing on differences in brain function and structure between stutterers and non-stutterers. The results suggest associated neural anomalies (Smith & Weber, 2017) and a neurological basis for the onset and persistency of stuttering (Chang). Although developmental stuttering begins during early childhood, few of these studies have targeted children. Importantly, children's brains are more likely to yield insights into the underlying causes of stuttering (Etchell, Johnson, & Sowman, 2014). This is because the differences found when comparing adults who stutter to adults who do not stutter may represent compensatory processes that have developed due to the neurological deficit underlying stuttering. However, as children have been stuttering for a shorter time, there is less likelihood that their brains have adapted to the stuttering. Consequently, children's brains are more likely to reveal the neurological causes of stuttering rather than the consequences.

Chang et al. (2015) were the first to conduct a neuroanatomical connectivity study in young children (3 to 10 years of age). Based on a sample of 37 children who stutter and 77 who do not, they found less connectivity in brain regions critical for skilled movement control among the children who stutter. They concluded that developmental changes in white matter may lead to recovery and further documented this association in a more recent study (see Chang et al., 2017). Most recently, Misaghi, Zhang, Gracco, Luc, and Beal (2018) investigated white matter abnormalities across major tracts underlying the neural network for speech production in a sample of 11 children who stutter and 11 children who do not. Their results indicated higher integrity and myelination in a white matter tract (the frontal aslant tract) on the right side of the brain was among the children who stutter than among those who do not. They interpreted these findings as indicating early right hemisphere compensation, however they did not find differences between the two groups of children in left hemisphere. These results tend to confirm the complex neurological underpinnings of stuttering from its onset. The gaps in the research literature can be bridged by prospective studies following children at risk of developing stuttering (i.e., first degree relative who stutters) throughout the time period when stuttering develops.

2.2.4 Factors associated with stuttering onset

Childhood stuttering can be considered a high frequency disorder in young children as between 5.19% (age span 3-5; Månsson, 2000) and 11.12% (age span 2-4; Reilly et al., 2013) experience stuttering for some period of time. A longitudinal study of 1619 Australian children (the ELVS study; Reilly et al. (2009)) found that stuttering occurred suddenly (over 1 to 3 days) for approximately half the children (49.6%) and involved whole word repetitions for the majority of the children (75%). Later, Reilly et al. (2013) documented that onset typically occurred between the age of two and four and that the majority of children started to stutter before they were 3.6 years old.

As noted by Smith and Weber (2017) and others, several developmental changes co-occur during the neurodevelopmental time window which stuttering occurs within. This is especially evident in language development, as stuttering typically begins at the time when children are combining words into short utterances—that is, in the third and fourth years of life (Packman, Code, & Onslow, 2007). The issue of whether stuttering is associated with weaker or stronger language skills has been widely debated (see Nippold, 2012). Ntourou, Conture, and Lipsey (2011) concluded that the language abilities of children who stutter may

differ subtly from those of children who do not stutter. However, Nippold (2012) argued that these reported differences only reflect variations in inclusion criteria and sampling strategies. In Reilly et al.'s. (2013) investigation stuttering in a community sample of children prior to onset of stuttering they found that the children who later developed stuttering had stronger language skills than those who continued to speak fluently on language assessment pre stuttering onset. Explaining the possible role of linguistic factors, MDP theory asserts that periods of rapid change in linguistic development have a destabilizing influence on the developing speech motor system.

There are inconsistent findings in relation to the temperamental characteristics of children who stutter. In their review of the literature, Kefalianos, Onslow, Block, Menzies, and Reilly, (2012) identified a number of recurring tendencies, including greater negativity in children who stutter (Eggers, Luc, & Van den Bergh, 2010; Ntourou, Conture, & Walden, 2013) and less inhibitory control among children who stutter (Anderson, Pellowski, Conture, & Kelly, 2003; Eggers et al., 2010; Walden et al., 2012). Nevertheless, as suggested by Smith and Weber (2017), there is no single psychosocial profile that characterizes children who stutter.

Importantly, as many existing studies have targeted children following the onset of stuttering, it seems possible that any observed differences between children who stutter and those who do not may be the consequence of stuttering rather than the cause. Reilly et al. (2013) made a valuable contribution to the debate by investigating the characteristics of children prior to the onset of stuttering. They found that gender, twinning status and mother's education level increased the risk of stuttering onset. While it is somewhat discouraging that these four factors explained only 3.7% of the total variation in stuttering onset, the inconsistency of findings is not surprising in light of the dynamic and individual pathways into stuttering proposed by MDP theory; Smith and Weber (2017) highlighted the heterogeneity in development in different domains. This involves that the group of children who stutter can show the full range of scores in domains of relevance to stuttering. Furthermore, children also differ in terms of factors that contribute to the development of stuttering.

2.2.5 Factors associated with recovery

One of the more fascinating features of stuttering is the sudden, unpredictable and spontaneous nature of recovery. According to MDP theory, recovery is explained with the development of neural connections that support a stable speech motor system. Most likely, this involves neural growth and connectivity changes in the left hemisphere that is relevant for speech productions (Smith & Weber, 2017).

Less than 10% of children recover during the first year after stuttering onset (Reilly et al., 2013; Ehud Yairi & Ambrose, 1999). However, recovery rates of approximately 70% have been reported (Yairi & Ambrose, 1999) as children grow: 71% by 5 years of age (Månsson, 2000), 65% by 7 years (Kefalianos et al., 2017) and 72% within four years after the onset of stuttering (age range at onset: 25–59 months; Yairi & Ambrose, 1999). Despite different percentages of recovery, there is consensus that the majority of young children who stutter do recover.

The factors associated with recovery include language skills: stronger language skills in girls at age 2 predicted recovery by age 7 (Kefalianos et al., 2017); steeper growth in syntactic production (mean age at assessment 57, 71, 83 months; Leech, Ratner, Brown, & Weber, 2017). However, Spencer and Weber-Fox (2014) reported no association with receptive or expressive language skills. With regard to gender, recovery has been associated with or being a boy (12 months post onset; Reilly et al., 2013), being a girl (Ambrose, Cox, & Yairi, 1997), as well as findings of no association with gender (Kefalianos et al., 2017). Recovery has also been linked to stuttering severity, in terms of more stuttering-like disfluencies at onset (Yairi et al., 1996), reduction in stuttering severity over time (Yairi et al., 1996) and lower mean ratings of stuttering severity after onset (Reilly et al., 2013). Heritability has also been found to be associated with recovery; for example, Dworzynski et al. (2007) found higher concordance rates among monozygotic twins in terms of recovered or persistent status at 7 years of age.

2.2.6 Factors associated with persistency

Over the school years, the prevalence of stuttering 1.44%, and by adulthood, the figure is 0.72% (Craig, Hancock, Tran, Craig, & Peters, 2002). In the literature on persistent childhood stuttering, there are some indications of poorer language skills in the group of children who persist to stutter measured at age 7 (Kefalianos et al., 2017) and 4 years after

stuttering onset (Ambrose, Yairi, Loucks, Seery, & Throneburg, 2015). The literature on temperamental characteristics is inconsistent; while Ambrose et al. (2015) found that parents perceived children whose stuttering persisted as more negative in mood, Kefalianos et al. (2017) found no differences in temperament. Dworzynski et al. (2007) suggested that children's own reactions, as well as the reactions of others, might be among the unique environmental influences that interact with genetic factors. This is interpreted to mean that, in combination with genes, certain temperamental characteristics such as emotionality and negative experiences can influence the pathway of stuttering. Erdemir, Walden, Jefferson, Choi, and Jones (2018) found slower articulation following negative emotions in children who persisted to stutter. This was interpreted to indicate potential contributions by a complex interaction between speech motor and emotional processes in recovery or persistency of stuttering. Presumably because of this complex interaction between factors, the occurrence and further development of stuttering remains a puzzle with many missing pieces. However, as summarized by Packman and Attanasio (2017) "there is an extensive body of scientific research that gives hope for our ultimate ability to unwrap the nature of stuttering, divest it of its mysteriousness and develop ways to treat it effectively" (p. 2).

3. The impact of stuttering

The International Classification of Functioning, Disability and Health (ICF; WHO, 2001) framework will be used here to explore how stuttering affects children, focusing in particular on impairment components and contextual factors. The ICF encompasses all aspects of human health and certain health-related domains and provides a scientific basis for understanding and investigating health and health-related states, outcomes and determinants (WHO, 2001). The classification's three main categories are (1) body functions and structures, (2) activities and participation and (3) contextual factors (environmental and personal). These components can be expressed either in positive terms, indicating non-problematic aspects of health and functioning (activities and participation), or in negative terms, indicating disability (impairment of body functions and structure, activity limitations and participant restrictions). Due to its foundation for understanding health-related states, the ICF will be applied to review studies of the health states of young children who stutter. In particular, Florian et al. (2006) argued that the ICF is a universal and relational model for understanding functioning and disability, involving particular relevance for the field of special needs education. The ICF is also applied in investigations of impact on school-aged and older children (see for instance Lankman et al., 2015).

3.1 What is the impact of stuttering?

Among adults who stutter, it is generally accepted that this involves more than disfluency, and that stuttering is a complex and far-reaching disorder (Shapiro, 2011). In his definition of stuttering, Shapiro (2011) has incorporated this psychosocial aspect of the disorder "... stuttering refers to individualized and involuntary interruptions in the forward flow of speech and learned reactions thereto interacting with and generating associated thoughts and feelings about one's speech, oneself as communicator, and the communicative world in which one lives" (p.12). Research has focused on the further effects of stuttering, often referred to as the *impact* of stuttering. In adults, this includes the effects of stuttering on social functioning (Craig, Blumgart, & Tran, 2009), mental health (Craig et al., 2009), attractiveness and romantic relationships (Van Borsel, Brepoels, & De Coene, 2011), self-esteem (Klompas & Ross, 2004), employment (Bloodstein & Bernstein Ratner, 2008; Klein & Hood, 2004) and education (Klompas & Ross, 2004). According to Klompas and Ross

(2004), quality of life is an important measure of the impact of communication disorders. In such studies, (Craig et al., 2009; Klompas & Ross, 2004; Yaruss & Quesal, 2004), impact is understood to refer broadly to the negative influence of stuttering on a person's quality of life, including such aspects as relationships (family, friends, school/work-related), communication-related challenges, negative thoughts and emotions related to stuttering and behavioral changes (e.g., avoiding social situations).

Evidence of the wide-ranging impact of stuttering in the adult population has prompted recent research on how stuttering affects young children. Based on results from the adult population, these studies have investigated whether stuttering is associated with reduced quality of life (de Sonnevile-Koedoot, Stolk, Raat, Bouwmans-Frijters, & Franken, 2014; Reilly et al., 2013); anxiety (Kefalianos, Onslow, Ukoumunne, Block, & Reilly, 2014; van der Merwe, Robb, Lewis, & Ormond, 2011); other children's reactions or changes in interaction with peers (Langevin, Packman, & Onslow, 2009, 2010); negative communication attitudes (Clark et al., 2012; Vanryckeghem et al., 2015); and when children become aware of stuttering (Boey et al., 2009; Ezrati-Vinacour, Platzky, & Yairi, 2001). Figure 2 illustrates the impairment and disability of the stuttering disorder (impact of stuttering) in relation to the ICF domains.

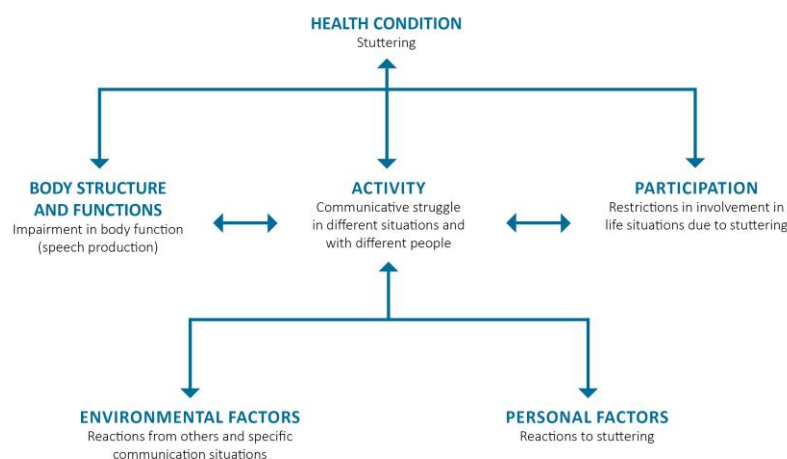


Figure 2. Stuttering and its impact (based on ICF domains).

3.1.1 Environmental factors

Environmental factors refer to the physical, social and attitudinal environment in which people live and conduct their lives (WHO, 2003). While the contribution of environmental factors differs across individuals, disability always entails an interaction between features of the person and features of the overall context in which the person lives (WHO, 2003). In relation to people who stutter, Starkweather (2002) highlighted cultural differences in how people react to stuttering. This creates either a safe and supportive environment or one where negative thoughts or emotions are more likely to arise as a result of negative listener reactions to stuttering (e.g., commenting, laughing, excluding).

In their investigation of attitudes among children who do not stutter, Ezrati-Vinacour et al. (2001) found that a majority of these children (68.8%) favored a puppet with fluent speech as against one with disfluent speech. Their interpretation of this finding was that young children tend to evaluate stuttered speech negatively. Investigating the social environment of children who stutter, Langevin, Packman, and Onslow (2009, 2010) reported that children who stutter experienced negative reactions from their peers, who showed impatience, teased them, made decisions for them, dominated play, ignored them, called them names and mimicked them or made fun of their stuttering. Yaruss, Coleman, and Quesal (2016) emphasized the importance of reactions to stuttering (their own and others’; see section 2.3.3) in terms of activity limitation and participant restrictions. In their study of the impact of stuttering on 20 school-aged children, they reported a strong correlation between the child’s own reactions to stuttering (OASES-S Section II) and activity limitation (OASES-S Section III) ($r = .73$) and participant restriction (OASES-S Section IV) ($r = .72$). However, potential associations between others’ reactions and activity limitation and participant restriction have not yet been investigated.

3.1.2 Personal factors

Personal factors include features of the individual’s life other than health condition (WHO, 2001). As measured by the OASES-S, children’s negative reactions were categorized as personal factors by Yaruss and Quesal (2016), who stated that reaction to stuttering “represents” the person’s coping style, and that this component helps to differentiate the

varied life experiences of individuals with the same health condition. These factors can also be considered part of the stuttering condition.

Emotional reactions

Among adults who stutter, anxiety in speaking-related situations is seen to result from negative experiences in communication situations throughout the life span (Iverach, Menzies, O'Brian, Packman, & Onslow, 2011). There is evidence that these experiences can occur even in young children in kindergarten (Langevin et al., 2009). Based on MDP theory, Smith and Weber (2017) proposed that children who stutter are likely to experience negative emotions when trying to regulate speech fluency, and that this emotional arousal may in turn destabilize the speech motor system. This can be understood as a negative loop, leading to further negative emotions. Studies investigating group differences between children who do and do not stutter indicate an association between reactivity and stuttering frequency (Choi, Conture, Walden, Jones, & Kim, 2016; (Jones, Choi, Conture, & Walden, 2014). In line with MDP theory, emotion reactivity and regulation can influence the stuttering pathway for some children. (Impairment of sensorimotor processes involved in speech production is necessary but not sufficient for stuttering to occur, and emotional reactivity and regulation is sufficient but not necessary.) For some children who stutter, then, emotional processes form part of a comprehensive account of childhood stuttering. Presumably, children with these general temperamental characteristics of reactivity and regulation will experience more emotional reactions to stuttering than others whose stuttering pathway is influenced by other factors (e.g., language).

Cognitive reactions

Speech-related thought and attitudes (communication attitudes) or cognitive reactions have been used to describe the negative thoughts and attitudes toward speech associated with stuttering. These two constructs differ in terms of specificity. As operationalized by Vanryckeghem and Brutten (2006), *communication attitudes* are general questions about the child's speech, such as "Do you like how you talk?" There is repeated evidence of differences in communication attitudes between children who stutter and children who do not (see Paper 1 for a review), indicating an association between stuttering and negative communication attitudes. Negative communication attitudes are also found in children with other communication disorders (e.g., voice and articulation disorders) (Luc & Brutten,

1990). As operationalized by Yaruss et al. (2016), *cognitive reactions* are negative thoughts that relate specifically to stuttering, such as “I can’t say things I want to say because I stutter.” As these questions relate specifically to stuttering, they are relevant only in assessing the negative thoughts of children who stutter.

Vanryckeghem, Hylebos, Brutton, and Peleman (2001) found a strong association between negative communication attitudes and negative emotions ($r = .89$) in school-aged children. Notably, the measure of negative emotions related specifically to items in the Communication Attitude Test (CAT), indicating that negative communication attitudes were associated with negative feelings but these results did not indicate the association between emotional reactions and cognitive reactions. Nevertheless, this may indicate a close association between negative emotions and thoughts related to stuttering.

Behavioral reactions

As well as causing negative emotions and negative thoughts, stuttering has also been shown to cause behavioral changes, including tension and struggle in the stuttering moment, avoidance of speaking situations and use of filler or starter words (Yaruss & Quesal, 2016). The most frequent behavioral reactions reported by Boey et al. (2009) were change in posture or attitude (26.0%), leaving or stopping talking (7.8%), head ticking (4.5%) and clownish behavior (3.2%) (in children younger than 48 months). Boey et al. (2009) interpreted these behaviors and others as indicators of awareness. For the majority of the children, these behaviors were present within one month after stuttering onset. Boey et al. (2009) also described that the behavioral reactions changed with age. For instance, stopping to talk or leaving the situation decreased with increasing age while asking for help increased with age. These results were interpreted by Boey et al. (2009) to indicate that experiences and the children’s cognitive and language developmental level were associated with their reactions to stuttering.

3.1.3 Body structure and function

Body functions refer to the physiological and psychological functions of body systems, and *body structures* are anatomical parts of the body (WHO, 2001). Of specific relevance to stuttering are the structures related to speech (e.g., neurological structures, larynx, articulators) and physiological functions associated with speech (e.g., respiration, phonation and articulation) (Yaruss & Quesal, 2016). In terms of body structure, research findings

(reviewed in X.X) indicate a neurological impairment among people who stutter; in relation to body function, people who stutter have difficulty in producing fluent speech (Yaruss & Quesal, 2016).

Studies of young children who stutter have investigated impairment of body function in terms of stuttering frequency (percent syllables stuttered; rating of how often the child stutters; see Millard, Paper 2); stuttering severity (often rated on a scale by parents or speech language pathologists (SLP); see de Sonnevile-Koedoot, Stolk, Rietveld, & Franken, 2015); or using the Stuttering Severity Instrument (Groner, Walden, & Jones, 2016; Tumanova, Conture, Lambert, & Walden, 2014)). Studies of older children have also used self-rated severity. Degree of stuttering severity as rated by SLPs and by children is known to be associated with children's experience of the impact of stuttering (Lankman, Yaruss, & Franken, 2015), indicating that body structure is associated with activity limitation and participant restriction in school-aged children.

3.1.4 Participation restriction

Participation restrictions relate to problems experienced by an individual in life situations (WHO, 2001). Based on the ICF Children and Youth Version (ICF-CY) (WHO, 2007), domains within this category considered relevant for young children who stutter include communication, learning, major life areas, community, social and civic life and interpersonal interactions and relationships. In young children who stutter, disorder-specific questionnaires assessing the impact of stuttering have revealed that parents perceive stuttering to affect the children's interactions and relationships and participation in specific activities (Langevin et al., 2010) (see Paper 2). Several of the above mentioned areas in the ICF-CY are included in general health-related quality of life (HRQoL) measures and studies that have compared children who stutter with children who do not stutter on such measures have concluded that stuttering does not have an effect on young children's HRQoL (see de Sonnevile-Koedoot et al., 2014; Reilly et al., 2013) (see Paper 2, section 1.2).

3.1.5 Activity limitation

Activity limitation refers to difficulties encountered by an individual in executing activities (WHO, 2001). For people who stutter, communication situations are especially difficult, and

limitations in activities may be associated with functional communication difficulties (Yaruss & Quesal, 2016). As reviewed above, research has extensively investigated the reactions of young children who stutter. However, the consequences of stuttering on the children's functioning have to a smaller extent been investigated. In school-aged children, the Speech Situation Checklist (SSC) assesses speech disruption in 55 speech situations (Brutten & Vanryckeghem, 2006), and communicative struggle is assessed by 15 questions in OASES-S (Yaruss et al., 2016). To the author's knowledge, there are no equivalent instruments for young children, other than one section in the newly adapted Overall Assessment of the Speaker's Experience of Stuttering—Caregivers (Parents and Kindergarten Teachers) (OASES-C) (Guttormsen, Yaruss, & Næss, 2018).

3.2 Measurement of impact of stuttering

As reviewed above, investigations of impairment and disability in young children have focused mainly on personal factors. The ICF, looks beyond the separate components and addresses the associations between them to understand health-states (see Figure 2). Yaruss and Quesal (2016) have used the construct *overall impact of stuttering* to encompass all of these components.

Smith and Weber (2016) highlighted the challenges of developing reliable and valid measures of emotional factors in young children, which also relate to the challenges of measuring how children are affected by stuttering. Parental reports have commonly been used in investigating the impact of stuttering (e.g., Langevin et al., 2010; Millard & Davis, 2016). However, as no study to date has targeted agreement between child and parent report by application of the same instrument for both groups, the level of agreement between children and parents remains unclear. In discussing agreement or consistency between child and parent reports, it is therefore considered useful to refer to the literature of HRQoL, as these measures address several of the same issues as disorder-specific measures of stuttering impact (e.g., emotional and social health and functioning) and several studies have investigated agreement in parent and child report.

3.2.1 Self-report

Discussion of self-reports has centered on children's cognitive and linguistic capacities. For the youngest children below the age of 5, Fekkes et al. (2000) stated that application of

proxies in measurement of HRQoL is necessary as the children themselves are unable to complete a questionnaire. For the five-year-olds, Varni et al. (2007) concluded that children in this age can reliably report on HRQoL with an age-appropriate instrument. This was based on their findings that the assessment differentiated groups of children with and without conditions affecting HRQoL. Rebok et al. (2001) on the other hand, found that children in between five and eight years had significantly more problems in understanding many basic health concepts than children older than eight years. On that basis, Rebok et al. (2001) argued that only children aged eight years or more have the requisite language and cognitive skills to report their QoL. As reviewed in Paper 1, self-report of communication attitudes has been documented to reliably differentiate between children who do and do not stutter.

3.2.2 Caregiver and child agreement

It is well documented that information provided by proxy report is not equivalent to that provided by the children themselves (Varni, Limbers, & Burwinkle, 2007), and it is increasingly acknowledged that the children's perspective on HRQoL is as valid as that of the parents (Eiser & Morse, 2001). Varni et al. (2007) analyzed the agreement in reporting among 8,591 children (aged 5–16) and their parents using the PedsQL™ (Varni, Seid, & Kurtin, 2001). They found moderate to good agreement between parents and children, and this increased with age. In general, agreement between self-report and parent report is known to depend on the HRQoL domains assessed; for example, in their review of 14 studies investigating agreement on HRQoL measures, Eiser and Morse (2001) found that consistency was higher for physical health and lower for social or emotional HRQoL. The authors related this to observability; while physical health is mostly directly observable (e.g., being unable to ride a bicycle because of injury or to attend school because of flu), social and emotional health may be observable (when the child expresses his/her feelings or thoughts) or not (when feelings and thoughts are retained or hidden).

Discrepancies between child and parent ratings may have several explanations. Children may be unable to report correctly on social HRQoL because they are unable to understand the questions or use a rating scale correctly, or they may be unable to recall relevant episodes. Equally, parents may lack insight into their child's social and emotional HRQoL, or they may evaluate aspects of HRQoL in a different way. HRQoL measures seek to investigate the

individual's life experience (Solans et al., 2008), regardless of the method applied. To be reliable reporters of their children's HRQoL, parents must have good insight into the child's experiences and must evaluate those experiences in the same way. This bears on the core challenge in utilizing parental reports; although parents frequently make inferences about their child's behavior in order to understand their needs, motives or limitations, their interpretations of the children's behavior and its causes may not necessarily be correct (Dix, Ruble, Grusec, & Nixon, 1986). It follows that self-report is generally preferred, and caregiver proxies are seen as a supplement rather than as a replacement. Nevertheless, caregivers are considered valuable informants in relation to children's behavior and potential changes due to stuttering by virtue of their ongoing observations across situations and time (e.g., responses to stuttering and reactions in the environment, struggle in communication situations and consequences of stuttering on behavior, mood and social participation).

3.2.3 Caregiver agreement

Two studies (Ntourou, Furlas, Marousos, and Paphiti, 2017, September; Wheeler, Fenton, and Millard, 2011, September) have investigated whether parents differ in their ratings of impact. Wheeler et al. (2011, September) found that parents who stuttered themselves rated impact higher than parents who had either no experience with stuttering or had recovered from stuttering, while Ntourou et al.'s (2017, September) found that mothers rated impact higher than fathers. As discussed in Paper 3 (section 1.2 and 1.3), there are several explanations of differences in perceptions of impact, both related to the informants (sensitivity, memory and interpretations of the children's behavior), the relationship between the child and informant and also the arenas the children are observed in. Karrass et al. (2006) advocated inclusion of reports by kindergarten teachers' in investigating emotionality in young children who stutter. This is equally important when measuring impact, as kindergarten teachers' education and observation of other children in the same developmental phase provide a strong theoretical and empirical basis for rating children's participation and activity foundation (Ibragimova, Granlund, & Björck-Åkesson, 2009). Additionally, other than parents, kindergarten teachers are the caregivers who commonly spend most time with the child. Prior studies have shown that parents ratings are stronger associated than parents and kindergarten teachers when reporting on children's behavior (see Paper 3, section 1.3).

4. Summary of studies and main findings

Two studies were conducted to explore how children are affected by stuttering and to investigate the reliability of caregiver report of impact. Study 1 was a meta-analysis of children's self-report of impact, and Study 2 was an empirical study of caregivers' reports on overall impact of stuttering in young children. The main characteristics of the two studies and the three associated papers are summarized in Table 1.

Table 1
Overview of the studies

Study	Design	Paper	Aim	Sample	Results
Study 1: Communication attitudes in children who stutter	Meta-analytical review of children's self-reports	Paper I: Communication attitudes in children who stutter: A meta-analytical review (Guttormsen, Kefalianos & Næss, 2015)	To summarize published and unpublished studies of communication attitudes in children who stutter (CWS), and to assess differences between studies.	18 studies involving children's self-reports of communication attitudes	Large difference in communication attitudes between children who stutter and children who do not. Age influenced the difference in communication attitudes, but gender did not.
Study 2: Caregivers' perceptions of impact of stuttering on young children	Cross sectional, quantitative and qualitative Parental report Caregiver report	Paper II: Parents' perceptions of the overall impact of stuttering in young children (Guttormsen, Yaruss & Næss, 2018).	To evaluate the parent questionnaire OASES-C. To investigate parents' perceptions of how stuttering affects young children and certainty when reporting on impact.	Parents perceptions of impact of stuttering in 38 young children (OASES-C)	The results indicate that the OASES-C is a suitable measurement tool. Parents perceived stuttering to have a moderate impact on young children and perceived themselves as certain in their ratings.
		Paper III: Caregivers' Perceptions of Stuttering Impact in Young Children: Inter-rater Reliability of Mothers', Fathers' and Kindergarten Teachers' Ratings (Guttormsen, Yaruss & Næss, 2018)	To investigate caregivers' perceptions of impact of stuttering in young children and agreement between caregivers in perceptions of impact.	Mothers', fathers' and kindergarten teachers' perceptions of impact of stuttering in 28 young children (OASES-C)	The caregivers perceived stuttering to affect the children similarly. Overall, the results indicated good consistency between caregivers. Findings also indicated disagreement on individual level.

The major reason for conducting a meta-analysis relates to the foremost advantage with this method: summarizing multiple studies including different persons, ages, settings and operationalisations of constructs enables generalization to different conditions (Shadish, Cook, & Campbell, 2002). Summarizing studies with different age group, settings and instruments, this study contributed the robust finding of significant differences in communication attitudes between children who stutter and children who do not. The large difference between the two groups indicates that children provided reliable responses to the existing instruments assessing communication attitudes.

The second study was driven by the absence of a comprehensive measure of overall impact for young children and the widespread use of caregivers' reports and the lack of research investigating the reliability of such reports. The study was an empirical investigation of how different caregivers perceived the impact of stuttering on young children. With this purpose, The Overall Assessment of the Speaker's Experience of Stuttering – Caregivers (Parents and Kindergarten Teachers) (OASES-C) was developed by adapting the version for school-aged children. The study contributed a number of novel findings. 1) Ratings of overall impact indicated that parents perceived stuttering in young children as associated with negative reactions to stuttering, communication difficulties, adverse effects on quality of life and limited knowledge of stuttering and stuttering treatment (Paper 2). 2) Parents perceived themselves as certain in their ratings of impact (Paper 2). 3) Overall, mothers, fathers and kindergarten teachers were had a good degree of agreement in their ratings of impact, although agreement varied across the four sections of the instrument. On individual level disagreement between a caregivers for a few children were revealed (Paper 3).

5. Methodological considerations

One of the two aims of this thesis was methodological: to investigate the reliability of assessment of impact of stuttering. In different ways, the studies included here address the assessment of impact, investigating self-report of communication attitudes across studies and measurement instruments (Paper 1); construct validity and internal consistency in parents' ratings of impact on OASES-C (Paper 2); and inter-rater reliability in caregivers' ratings of impact on OASES-C (Paper 3). The methodological focus of this extended abstract reflects the focus on assessment of impact throughout the thesis.

Research commonly involves choices regarding method, design, sample, instruments and analysis. All of these methodological choices influence the validity of the inferences drawn from the results and therefore warranted careful consideration when planning and conducting this research. The first sections of this chapter (5.1-5.3) elaborate and discuss the crucial methodological choices made throughout the research process in respect of both studies. As Study 2 relates to stuttering in young children in general, including the primary studies in the meta-analysis, the choices in Study 2 were necessarily more focused. The consequences of these choices for validity will be discussed in the context of Shadish et al.'s (2002) well-established typology (section 5.4). Additionally, the discussion of validity will highlight issues for investigations of impact in relation to children, expanding on discussion in the articles. Construct validity will be examined as a crucial methodological issue for such investigations and therefore for this project as a whole. Reliability will be discussed in section 5.5 as the other main indicator of quality of measurement tools (Stolarova, Wolf, Rinker, & Brielmann, 2014). The chapter closes with a discussion of the project's main limitations. These critical discussions of the methodological nuances of the studies supplement the papers themselves.

5.1 Methodological choices

5.1.1 Choices related to inclusion criteria

As highlighted by Kefalianos et al. (2017), discrepancies in studies of the incidence of stuttering may reflect the inclusion criteria applied (age and time since onset), how stuttering

is operationalized and how participants were recruited. These methodological choices can also influence the results of investigations of impact. Therefore these choices will be justified and discussed in the paragraphs below.

Age

In Study 1 and Study 2 different age groups were included according to the aim of the studies and the different data collection instruments applied. Studies of the impact of stuttering have mainly targeted children above the age of three when using self-reports (e.g., Brce & Vanryckeghem, 2017; Clark et al., 2012; Vanryckeghem, De Niels, & Vanrobaeys, 2015) and children above the age of two when using parental reports (e.g., Langevin et al., 2010; Millard & Davis, 2016). The two studies included in this thesis also adopt this approach; three years was set as the lower age limit in Study 1 because children develop their ability to talk about mental states (Bretherton & Beeghly, 1982) and to use and understand language about the self (Brownell, Ramani, & Zerwas, 2006) between the age of two and three. In Study 2, however, children from the age of two were included because of aim of the study and the application of parent proxies to investigate impact. However, age can also be a challenge in such reports, as caregivers may have less insight into these younger children's thoughts precisely because they lack the ability to talk about their inner states (for discussion, see section 4.2, Paper 3). Thus, the developmental level of two- to three-year-olds makes investigation of their inner states challenging, regardless of method.

Time since onset

Studies of stuttering in young children vary in time since onset (TSO) criteria. Of the primary studies included in Study 1, the majority of the studies did neither applied a specific amount of TSO as an inclusion criteria, nor presented these characteristics (e.g., Abbiati et al., 2012; Brce & Vanryckeghem, 2017; Clark et al., 2012; Vanryckeghem & Brutten, 2006; Vanryckeghem et al., 2005; Węsierska & Vanryckeghem, 2015). However, as highlighted by both Tumanova et al. (2014) and Clark et al. (2012), TSO is an important aspect to include in investigations to understand individual differences and how quickly negative communication attitudes develop. In Study 2, to investigate children with a wide range of stuttering experiences, children were included one month after onset. This minimum of one month since stuttering onset reflects the need for parents to have time to observe impact before answering the questionnaire, and as they were asked to report on the impact of stuttering

over the previous month. Naturally, the two characteristics age and TSO are closely associated (Tumanova et al., 2014).

Operationalization of stuttering

None of the studies included in this thesis applied the common criterion of a minimum 3% of syllables stuttered in a speech sample (Conture, 2001). This was because such a cut-off can wrongly exclude children who stutter, as the percentage of syllables stuttered may be at the margins of this criterion (Tumanova et al., 2014). In the review of communication attitudes, the differences within the field in regards to operationalization of stuttering became evident as there were noticeable differences in operationalization and measurement of stuttering—for instance, criteria for inclusion in the group of children who stutter included frequency of stuttering (e.g., Clark et al., 2012), types of disfluency (e.g., Bernardini, Vanryckeghem, Brutten, Cocco, & Zmarich, 2009), assessment and diagnosis of stuttering (e.g., Kawai, Healey, Nagasawa, & Vanryckeghem, 2012) and treatment status (e.g., Beilby et al., 2012). As Packman and Attanasio (2017) pointed to, reaching consensus in operationalization of stuttering would mean that research could be conducted with agreement of what is being measured. Consequently, such discrepancies in operationalization can prevent direct comparison across studies, and this may also threaten the validity of Study 1. However, as features of stuttering challenges consensus in operationalization and defining stuttering and as there to date are no consensus (Packman & Attanasio, 2017), it was decided to include studies with different operationalization of stuttering.

To ensure that only children who stutter were included, Study 2 employed several measures of stuttering (%SS from a speech sample and severity ratings) and stuttering was confirmed by three different sources: severity ratings by both parents for the previous week and confirmation of stuttering by either the project leader (undersigned) or the SLP treating the child.

5.1.2 Choice related to recruitment procedures

Recruitment procedures in Study 2 were selected to ensure as large a sample as possible. Some of these recruitment strategies were similar to those of the primary studies in Study 1; some related specifically to the health care system in Norway; and some were adopted to reach out broadly across the whole country. First, it was decided to collect data throughout

the whole PhD period after the study was approved by the Norwegian Centre for Research Data (from January 2015 to February 2018). Secondly, close collaboration with important organizations or institutions was prioritized (e.g., national interest organization for people who stutter, the national organization for SLPs and departments of fluency disorders at national service centers for special needs education) because of their networks of parents and SLPs dealing with children who stutter. Information about the project was also distributed to kindergartens in the capital (by physical visit, email or telephone) and to SLPs working in the municipality or in private practice in the four counties nearest the capital. To reach out nationally to parents of children who stutter, information was disseminated through Norwegian media, including interviews in a parenthood magazine and health care web sites and on two nationally broadcast radio programs. This broad recruitment strategy was designed to ensure a representative sample consisting both of children who did and did not receive treatment.

5.1.3 Choices related to data collection methods and measures

Self-reports of communication attitudes and caregiver reports of impact

In Study 1, communication attitudes reported by the children themselves was summarized while caregiver report was applied to investigate impact in Study 2. Although Study 1 and Study 2 employ different methods to assess the impact of stuttering, the choices of methods are not seen to conflict, as the different aspects of impact targeted in the two studies directed the choice of methods. On the assumption that children develop negative thoughts about their speech because of stuttering, the findings in Paper 1 indicate that both young and older children can reliably report on communication attitudes using age-adjusted questionnaires. However, measurement of overall impact of stuttering involves reporting on prior experiences and how one's life is affected by stuttering, and as young children have been shown to struggle with understanding, expressing and recalling their internal states (Brownell, et al., 2006; Rebok et al., 2001), caregiver reporting was considered the most appropriate method.

The choice of a caregiver proxy questionnaire posed several challenges. First of all, it had a significant influence on the inferences that could be drawn, as the results indicate only impact as perceived by caregivers and not impact as experienced by the children. A second major challenge related to measurement of internal states—in particular, of communication attitudes or cognitive reactions to stuttering. Caregivers' insights into children's internal

states reflect the extent to which the children show their feelings or express their thoughts through behavior that can be interpreted. Additionally, insight may also be dependent on parent's sensitivity, memory and interpretation of the children's behavior (Boey et al., 2009). Consequently, caregivers might differ in their views of how children are affected by stuttering and in the extent to which their answers reflect the children's own experience. Nevertheless, this approach was chosen because understanding the differences between caregivers' perceptions and insights into children's internal states can help to enhance the reliability of such assessment procedures.

Parents' insights into the impact of stuttering

Parents were asked to rate their level of certainty when answering questions on a 5-point Likert-type scale, ranging from *not certain at all* (1) to *very certain* (5) (see Table 4, Paper 2). On the assumption that parental reporting should be used only if parents feel reasonably certain, this measure was included to investigate the appropriateness of parental reporting in assessment of overall impact.

Estimations of accuracy within the interpersonal perceptual domain, as in the present study, can never be absolute (Biesanz et al., 2011). For that reason, the reliability of self-perceived certainty can be difficult to investigate, and studies of this kind proved difficult to find. Investigating the association between estimated accuracy and actual accuracy of such judgements, Biesanz et al. (2011) found that self-evaluation of accuracy was associated with actual accuracy, and that people seemed to be aware of fluctuations in accuracy across different settings. Ratings in the different sections of OASES-C indicated less certainty when reporting on children's inner states (e.g., attitudes, knowledge, awareness and cognition in Sections I and II), indicating that self-ratings of certainty offer one suitable method of assessing the appropriateness of parental reporting. Good inter-rater reliability and the low percentage of IDK responses also supported the appropriateness and reliability of caregiver reports. Importantly, none of these results indicates whether parents' responses are identical to those of the children.

5.1.4 Choices related to OASES-C

Study 2 was a comprehensive investigation of caregivers' perceptions of how young children can be affected by stuttering. The OASES instrument for school-aged children was adapted

to provide a caregiver proxy version (see section 2.2.1 in Paper 2 for a description of this process) that investigates the overall impact of stuttering in young children. There were several reasons for choosing to use OASES to measure impact. First, the OASES instrument defines impact of stuttering in terms of a recognized international framework (the ICF). As it is often unclear why certain items are included in measures of impact while others are not (Yaruss and Quesal, 2016), the ICF provided a solid foundation that accounts for the included items and constructs. The universality of the framework also provides a broad perspective on the specific health aspects of stuttering. Given the instrument's strong theoretical foundation and good reliability and validity (Yaruss & Quesal, 2016), it was considered useful for the purposes of Study 2 to develop a caregiver proxy version of the instrument for young children. The association between the instrument and the framework was considered pertinent because the application of a theoretical framework for understanding the impact of stuttering differentiates OASES-C from other instruments assessing impact in young children.

A closer look at the association between OASES items and ICF components (see p. 5 in Yaruss & Quesal, 2016) reveals that the majority of the OASES-C items are associated with the ICF, even though several are not. This becomes clear in Table 2, where aspects of impact described in the manual without reference to an ICF component are termed *other aspects*.

Table 2

Overview of OASES-C content and ICF components

OASES-C section (N items)	ICF component and OASES-C items (N items)
Section I (15)	Body function and structure: perception of impairment (4) Other aspects: Knowledge of stuttering and treatment (5); attitudes and feelings towards speech, stuttering and treatment (6)
Section II (20)	Personal factors: emotional, behavioral and cognitive reactions to stuttering (20)
Section III (15)	Environmental factors and activity limitation: difficulties in different communication situations (15)
Section IV (10)	Participation restriction: how much stuttering hinders involvement in different life situations (5) Other aspects: impact on quality of life of stuttering and other people's reactions (5)

As indicated in Table 2, 16 items in OASES-C are not considered to be closely associated with the ICF. However, the most obvious discrepancy is that the ICF addresses only health and states of health while OASES-C focuses specifically on the consequences of stuttering for health. This reflects the ICF's goal of being applicable to all, irrespective of health condition, and to describe health irrespective of health condition (WHO, 2002). In contrast, OASES-C was developed to assess the consequences of stuttering for the communication and life experiences of young children who stutter (see Paper 2), and only these aspects are included in OASES-C. Another difference between the instrument and the framework is that OASES-C includes impact on quality of life. This is not visualized in the ICF model, even though its components can be seen as facets of quality of life (McDougall, Wright & Rosenbaum, 2010). McDougall et al. (2010) advocated for inclusion of QoL in the ICF, as the ultimate goal of good health and functioning is enhanced QoL. A third discrepancy relates to the development of a caregiver proxy version as the OASES-C involves, as this assesses caregivers' perceptions of impact rather than those of the speakers themselves (as assessed in other OASES instruments).

5.1.5 Choices related to analysis and statistical approach

Missing data

Missing data may indicate that one does not know or does not wish to answer the question (Field, 2003). In Study 2, two choices were made in relation to missing data; IDK responses were treated as missing, and participant means were imputed for the missing data. The reason for treating IDK responses as missing was that all participants were "forced" to respond to all items in the electronic version of OASES-C. (An error message appeared stating that respondents could not finish until all items were completed.) As there was no option to omit any item, IDK responses were taken to indicate the respondent's unwillingness to answer or their lack of insight.

Handling missing data by imputing a participant average aligns with Schafer and Graham's (2002) recommendations for good reliability. While complex procedures related to missing randomness could have been applied to handle missing data, Shrive, Stuart, Quan, and Ghali (2006) argued that the simpler procedure of imputing a participant mean is appropriate for data sets of this kind. In their investigation of different imputation methods for missing

values on a questionnaire assessing depression, they found that imputation of participant mean was one of the methods that produced the most favorable results for data sets in which 10% of values were missing. Although missing data constituted less than 10% of parents' ratings (see Table 4, Paper 2), it cannot be ruled out that this choice of procedure for handling missing data may have influenced the results.

Investigation of normal distribution

For small samples in particular, it is important to investigate whether the data are normally distributed, and several procedures can be applied for this purpose. In Papers 2 and 3, skew and kurtosis were converted to z-scores for significance testing. These values were then compared to the values one would expect if skew and kurtosis differed from 0 (see Table 4 in Paper 2 and Table 2 in Paper 3). However, as small samples may lack the power to detect violation of normal distribution (Field, 2013), it can be argued that several assessments of normality should have been conducted. For instance, scores in the sample could have been compared to a similar set of scores that were normally distributed, using the Komogorov-Smirnov test or the Shapiro-Wilk test. This may have produced different results, as these tests differ in terms of how normal distribution is assessed and in their power to detect differences (Field, 2013). Nevertheless, it was decided only to assess normality using z-scores, as this provides information about how far the values are from 0.

Intra-class correlation

Intra-class correlation (ICC) was chosen for the analysis of agreement between caregivers' ratings of impact because it reflects consistency and agreement between measures of the same class (i.e., ratings of the same children) for continuous data (Field, 2013). Stolarova et al. (2014) noted that linear correlation analysis (Pearson's r) is frequently used in investigations of inter-rater reliability, where ICC analysis would be more appropriate. This is because linear correlation provides information about the relation between two variables but not about the degree to which caregivers' exact ratings are similar. It follows that consistently lower ratings by one group can produce a strong correlation coefficient and may be interpreted as indicating good agreement (Stolarova et al., 2014). However, such tendencies are not hidden by ICC analysis of absolute agreement, which reflects both caregivers' ranking of impact and the scores' absolute values.

ICC is based on the assumption of normal distribution (Shrout & Fleiss). Despite non-normal distribution tendencies in the data in Paper 3, ICC was considered the most

appropriate method of analysis because the majority of ratings were normally distributed (27 of 30) and because these non-normal tendencies were not considered gross violations of normality (see Hallgren, 2012). However, in interpreting the results, it should be kept in mind that the non-normal distribution in Section IV and the overall score may have influenced the results.

As there are different types of ICC analysis, whose application can produce different results, the options were discussed with a colleague specializing in ICC analyses and an expert in statistics in order to minimize the risk of incorrect use. The appropriate ICC was selected on the basis of guidelines developed by Shrout and Fleiss (1979) and McGraw and Wong (1996), with further explanations by Hallgren (2012).

First, the choice of one-way or two-way analysis relates to whether the subjects are rated by the same raters (two-way) or by different raters (one-way). All of the children were rated by their mother, their father and their kindergarten teacher. Consequently, raters differed from child to child (except for two children that were siblings). However, as the raters represented a group of caregivers and all children were rated by all three caregivers (i.e., their mother, their father and their kindergarten teacher), a two-way analysis was applied. Secondly, average measures were chosen since all of the children were rated by all three raters (i.e., mothers, fathers and kindergarten teachers) and the main results involved the average ratings by the three groups (shown in Table 2 in Paper 3). Average measures compared to single measures generally produce more reliable measurements (Hallgren, 2012). In our results, this was evident as there was a large discrepancy between estimates with single and average measures. Even though average measures were considered to be the most appropriate ICC type to apply, McGraw and Wong (1996) argue that one should include both types if there is a large discrepancy between the estimates. However, as choices concerning ICC types require knowledge beyond what can be expected of the readers, we chose to only report on the appropriate ICC type to make the results as available and easily interpretable as possible for the readers.

A mixed effects model was applied as the raters were not randomly sampled (i.e., the raters were not randomly recruited from a sample of raters, but rather specifically chosen due to their caregiver role for the child that was included in the study) and because the aim of the

study was to investigate the reliability of the raters (Hallgren, 2012). Both consistency in caregivers' ratings (i.e., agreement in rating of the children; the children's relative scores) and absolute agreement (i.e., consistency and agreement in the rating values) was reported on.

Inclusion of qualitative analyses

One of the main strengths of qualitative data is the insight provided into the inner experience of informants (Corbin & Strauss, 2008). As Study 2 was an initial investigation of caregivers' perceptions of overall impact in young children, it was considered pertinent to include qualitative descriptions of how they perceived impact in order to enrich the quantitative impact ratings. As reviewed in Flick (2006), there are several ways of combining quantitative and qualitative data, depending on the research aims and questions.

To meet the papers' differing aims, the two data sources were integrated in different ways. In Paper 2, the qualitative examples of impact supplemented the quantitative ratings to gain a more in-depth understanding of impact as perceived by the parents of children who stutter. Analysis of the consistency of descriptions and sections was conducted to investigate construct validity. In particular, the qualitative data complemented the quantitative findings by accounting for some of the tendencies in the ratings, such as the frequency of IDK responses and possible irrelevant items.

In Paper 3, the qualitative data were used to facilitate the interpretation of agreement between caregivers' impact ratings. The analysis of agreement and disagreement among caregivers' descriptions of impact involved a systematic comparison of descriptions for each child in line with Richards' (2005) dominant patterns analysis. The purpose of the analysis was to explore agreement at the individual level and potential explanations for variations in caregivers' ratings, complementing the quantitative findings of group agreement tendencies.

5.2 Validity considerations

Validity refers to the approximate truth of an inference (Shadish et al., 2002). Whether the interpretation of the evidence holds across persons and settings is an ongoing empirical question, and validity is therefore an evolving property (Messick, 1995). To evaluate and discuss validity in quantitative studies, Shadish et al. (2002) developed a typology of four main forms of validity: internal validity (of causal inferences), construct validity (of

inferences drawn from indicators to constructs), statistical conclusion validity (of statistical inferences) and external validity (of inferences across persons, settings, measurements and treatments). Threats to validity explain why inferences concerning covariance, causation, constructs and generalizations may be partly or completely wrong (Shadish et al., 2002). This typology informs the discussion of possible threats to the validity of inferences drawn in this project.

5.2.1 Construct validity

Construct validity relates both to the understanding of constructs and the assessment of constructs (Shadish et al., 2002). In this study, different instruments and methods were used to assess the impact of stuttering; while all the primary studies in Study 1 used self-reporting, Study 2 employed parental reporting. In both studies, abstract constructs such as thoughts, attitudes, feelings were operationalized as concrete indicators of impact (for examples, see section 4.4 in Paper 1).

Definition of communication attitudes and impact

Shadish et al. (2002) stressed the importance of construct explication or definition for construct validity; only when a construct is thoroughly defined can one evaluate whether the assessment is too narrow to include all relevant aspects of the construct (construct underrepresentation) or too broad, including irrelevant or confounding constructs (construct overrepresentation). The instruments used to assess communication attitudes and impact of stuttering differ in terms of definitions provided and how specific those definitions are.

Millard and Davis (2016) specified goals to reduce overall adverse impact on a child's life. Their operationalization of the construct of impact in the Palin Parent Rating Scale (Palin PRS) incorporated avoidance, poor communication skills, diminished self-confidence, bullying, denial of stuttering and negative emotions. According to Yaruss and Quesal (2016), OASES measures the impact of stuttering in four areas: perceptions of impairment, reactions to stuttering, functional communication difficulties and impact of stuttering on quality of life (p. 35). Investigating communication attitudes, Vanryckeghem and Brutten (2006) (p. 2) stated that negative attitudes toward one's own speech stem in part from the belief that speech is difficult, but they provided no definition of communication attitudes in the manual

accompanying the Communication Attitude Test for Preschool and Kindergarten Children Who Stutter (KiddyCAT).

In part because of the lack of definitions and agreement on the content of constructs, instruments assessing impact and communication attitudes differ in content and comprehensiveness. As discussed in Paper 1 (section 4.4), the Communication Attitude Test (CAT; Brutten, 1985) measuring communication attitudes in school-aged children is more comprehensive than the version for kindergarten children (KiddyCAT); factor analyses revealed three underlying factors for the CAT (Dekort, 1997), and one for the KiddyCAT (Clark et al., 2012) (see section 4.4. in Paper 1 for a discussion). Comprehensiveness and construct dimensions also differ across instruments assessing impact; for example, the Palin PRS employs 7 questions to assess impact while the OASES-C assessment involves 60 items. One might question whether all aspects of impact are covered by less comprehensive measures of communication attitudes and impact or if the construct is underrepresented. In the case of more comprehensive instruments, one might ask whether irrelevant constructs have been included. No study known to the author has investigated the correlation between instruments and whether they assess the same construct.

Instrument specificity in measuring impact

In disorder-specific instruments assessing impact, there are several threats to construct validity. These are of relevance to both studies here, as OASES-C, OASES-S and OASES-T were included in both. First, as the items in disorder-specific instruments focus on the negative effects of stuttering, they cannot be considered neutral. As Langevin et al. (2010) argued in their discussion of items in the ISPP, “as it is likely that there would be few positive effects of stuttering, the questions were designed to measure the presence or absence of negative impact and to learn more about the nature of any negative impact” (p.410). In OASES-C, for instance, parents are asked to rate how much stuttering gets in the way of specific activities, and how negatively their children’s lives are affected by stuttering. The response categories range from *not at all* to *completely*. While no prior study has reported positive consequences of stuttering in young children, such formulations can bias results, as they indicate an expectation of stuttering’s negative impact. Secondly, the specificity of items poses a challenge, as caregivers are asked to report how stuttering affects different aspects of the children’s lives. Such questions presume that caregivers can understand the complex causal mechanisms behind the child’s behavior, thoughts and feelings. For instance,

factors other than stuttering may influence a child's well-being. With particular reference to stuttering, Guitar (2014) highlighted the potential influence of co-occurring life events on stuttering onset and development. In the case of a child who starts to stutter and simultaneously experiences extensive events, it can be challenging to investigate the impact of stuttering in isolation from other influential factors.

General HRQoL instruments (see discussion in section 1.2, Paper 2) have been used to measure various aspects of the impact of stuttering, along with instruments assessing communication attitudes (e.g., KiddyCAT, CAT and A-19 Scale; see Paper 1). By asking more neutral questions (e.g., yes/no answers to questions indicating presence or absence of negative communication attitudes), these general instruments overcome the validity threats that challenge disorder-specific questionnaires. They also facilitate comparison across different groups of children; for instance, children who do not stutter may have negative thoughts related to speech (as measured in KiddyCAT, CAT and A-19 Scale), but children who do not stutter are not expected to have negative reactions to stuttering (as measured by OASES-S&T). It follows that differences between children who stutter and those who do not may simply reflect differences in presence or absence of stuttering rather than in the given construct (e.g., communication attitudes) when investigated with disorder-specific instruments. For that reason, inclusion of the study using OASES-S&T to compare children who do and do not stutter (Beilby, Byrnes, & Yaruss, 2012) may have biased the results of the meta-analysis.

Conversely, the challenges related to disorder-specific instruments can also be seen as strengths because these instruments facilitate assessment of the specific perceived impact of stuttering on children's lives. As more general HRQoL assessment may not be sufficiently specific to determine the impact of stuttering (de Sonnevile-Koedoot et al., 2014), the children's treatment need may remain unknown if such assessment instruments are applied. Furthermore, the specificity of disorder-specific instruments makes them more sensitive to the effects of treatment; clinically, disorder-specific instruments can efficiently assess the aspects relevant to the disorder (Solans et al., 2008).

Construct validity of OASES-C

The underlying universal theoretical framework for understanding health that is common to all OASES instruments increases the likelihood that all relevant aspects of impact will be included, so strengthening construct validity. As discussed in section 5.1.4, OASES-C deviates from the framework in several ways. However, as the ICF is a framework for understanding health states in general, discrepancies between the instrument and the framework do not necessarily undermine the instrument's construct validity. Rather, some deviations from the framework may serve to ensure construct validity in measuring impact in young children, as in the inclusion of quality of life issues documented in several prior studies as impacts of stuttering (e.g., Langevin et al., 2010, Ntourou et al., September 2017). Additionally, while the OASES-C focus on the specific consequences of stuttering deviates from the general ICF perspective, it reflects the construct *impact of stuttering* as applied by Langevin et al. (2010) and Millard and Davis (2016).

Nevertheless, there are several threats to the construct validity of OASES-C. First, the adaptation of OASES-S to OASES-C may have reduced the questionnaire's construct validity, as some of the items related to knowledge in Section 1 may be less relevant for many of the younger children (see section 4.4, Paper 2). For older children, knowledge of stuttering and stuttering treatment may be associated with less severe impact, as it can contribute to positive feelings and thoughts (e.g., being in control, predictability, being seen and helped). However, as indicated by parents' qualitative responses (Paper 2), lack of knowledge may indicate low awareness or experience of stuttering and treatment rather than more severe impact. The fact that impact ratings were most severe in Section I and did not correlate with the other sections may indicate that some of the items in Section I assess irrelevant construct(s). The potential inclusion of irrelevant constructs may have biased the results, as the estimate of overall impact may have been less severe. This was commented on in Paper 2, and it will be crucial to investigate this issue further in validating OASES-C.

Construct validity can be investigated by analyzing an instrument's correlation with other instruments measuring the same construct or by assessing the instrument's ability to differentiate between two groups (Kleven, 2002). Given the specificity of OASES-C and in the absence of any other validated instruments assessing impact in Norwegian, construct validity was investigated by analyzing parents' descriptions and examples of behavior indicating impact (see Paper 2, section 3.2). These descriptions provided valuable knowledge

about parents' perceptions of items, including what they perceived the instrument to assess. However, the data did not clarify whether the indicators related to the theoretical construct *impact*.

5.2.2 External validity

External validity refers to the generalizability of results—that is, the extent to which inferences hold across variations in conditions (e.g., person, setting, age) (Shadish et al., 2002). The two studies here differ in their assumptions concerning external validity because of the different methods applied.

Generalizability of meta-analysis findings

Because meta-analyses examine studies involving different conditions and large samples, their findings are considered more precise and generalizable than those of primary studies (Matt & Cook). A larger sample includes more individuals from the targeted population and therefore provides an estimate that is more representative of the whole population (Borenstein, Hedges, Higgins, & Rothstein, 2009). Secondly, the inclusion of multiple samples involving a large number of diverse persons and settings makes it possible to generalize to broader classes than in primary studies (Matt & Cook, 2009). For instance, the results in Study 1 can be generalized across countries and cultures by virtue of the diversity of the primary studies.

Publication bias

Threats to the external validity of meta-analyses relate to the sampling of studies. In particular, as studies with a significant effect size are more likely to get published, they are also easier to find (Shadish et al., 2002). This difference in effect sizes between published and unpublished studies and the likelihood of including more published studies is referred to as *publication bias*, and this is considered a significant threat to the external validity of meta-analyses (Borenstein, Hedges, Higgins, & Rothstein, 2009). To minimize publication bias here, studies that had not been published in peer-reviewed academic journals (grey literature; Rothstein & Hopewell, 2009) were included in the meta-analysis.

One argument against the inclusion of grey literature in meta-analyses is lack of certainty about the relative quality of unpublished studies, which may not have been peer-reviewed as

thoroughly as published manuscripts (or at all) (Rothstein & Hopewell, 2009). Given that the majority of studies published on communication attitudes were written by the copyright holders of the instruments, the inclusion of grey literature in Paper 1 was considered to be of particular concern. The potential threat relates to the commercial reality that reporting an instrument's ability to differentiate between people who stutter and people who do not may be in the copyright holders' interest, as this may indicate good construct validity. It was therefore considered particularly important to identify all studies on the topic, including those by researchers with no commercial interest in the instruments. Notably, no copyright holder of the applied instruments was among the authors of the two papers that reported non-significant results (Abbiati et al., 2012; Devore, Nandur, & Manning, 1984) (for details see Figure 2, Paper 1). However, as three other studies (Clark et al., 2012; Jelčić Jakšić, 2012; Leahy & Loftus, 1998) by non-copyright holders reported significant differences between groups, it seems unlikely that biased inferences were drawn. The asymmetrical funnel plot and the trim and fill analysis indicated that publication bias may have influenced the size of the estimated effect, but not in the hypothesized direction, as the adjusted effect size was larger than the estimated effect size.

Representativeness of sample

As similarity between the sample and targeted population increases, one can also expect the validity of generalization to increase (Lund, 2002). As the whole population of children who stutter cannot be estimated, sample representativeness can be assessed by comparing child characteristics in the present sample to studies that included large samples of children who stutter, or to relevant meta-analyses. A comparison of sample characteristics in Paper 1 to prior studies by Yairi and Ambrose (1999), Månsson (2000) and Reilly et al. (2013) reveals that the high number of boys included may have reduced representativeness and therefore the generalizability of results in Study 2. However, as both Papers 2 and 3 address caregivers' perceptions of impact, one can argue that the representativeness of these informants is crucial for generalizability of the results. For instance, gender distribution has not been found to affect inter-rater reliability among caregivers (Duhig, Renk, Epstein, & Phares, 2000). As highlighted in Paper 3 (section 1.3), fathers' involvement in childcare may be one factor that influences inter-rater reliability among caregivers because more equal involvement and time spent in childcare is likely to lead to more similar observations of the children. Welfare systems in countries may have an influence on fathers' insight into impact of stuttering, as paid paternal leave associated with later engagement in childcare (Haas &

Hwang, 2008) and more emotional attachment to the children (Almqvist, Sandberg, & Dahlgren, 2011). As Norway is one of few countries with such social policy, agreement between Norwegian parents may be higher than for other countries where fathers do not have paid paternal leave where parents' involvement in childcare is more divergent.

5.2.3 Statistical conclusion validity

Small sample sizes minimize statistical power and the likelihood of significant results (Shadish et al., 2002). Many studies of stuttering, including Study 2 and some of the studies included in Study 1, are hampered in this regard. As low statistical power in primary studies increases the risk of rejecting true hypotheses (Shadish et al., 2002) but does not bias effect sizes in meta-analyses (Valentine, 2009), synthesizing results is especially important in fields where study samples are small. The range and mean of sample sizes in Study 1 ($M = 64.33$, $SD = 39.97$, range 5–149) indicate that studies in this area differ in statistical power, and while the sample in the empirical study here is not the smallest, it is considerably below the mean sample size.

As highlighted in the limitations of Papers 2 and 3, the sample size in Study 2 limited the options for analysis. It also affected the precision of results in Paper 3, as CI range varies from X to X (Table 4). Koo and Li (2016) specifically recommended a minimum sample size of 30 and a minimum of three raters. While the sample presented in Paper 3 has a sufficient number of raters, the sample size is slightly below the recommended minimum ($N = 28$). In addition to sample size, the width of CIs may also have been influenced by the distribution of data or by the study design (Ionan, Polley, McShane, & Dobbin, 2014). As ICC was applied to data with a limited number of values (average rating on a 5-point Likert-type scale), and as some sections for some caregiver groups were non-normally distributed, this may have affected the width of confidence intervals. In general, the uncertainty associated with the results affected the validity of statistical inferences.

A threat to statistical conclusion validity that is specific to meta-analyses is selective outcome reporting. This occurs where authors of primary studies have applied several measures of the same construct but only report the measure that produced statistically significant results (Valentine, 2009). This was not the case in Abbiati et al. (2012), as both measures were reported. However, as two measures were applied, and the two measures

produced different results—non-significant findings when assessing communication attitudes with KiddyCAT and significant differences between the groups when measuring communication attitudes with PASS—the choice of measures to include in the meta-analysis affected the results.

5.3 Reliability

Test reliability refers to the accuracy or precision of a measurement procedure (Thorndike & Thorndike-Christ, 2014). Measures of the reliability of assessment tools include internal consistency (the extent to which items in a given test cohere), test-retest reliability (consistency in answers across time) and inter-rater reliability (consistency across raters). The widespread assessment of internal consistency may relate less to its importance as a measure of reliability (Cicchetti, 1994) than to the fact that this measure does not require additional data collection.

The primary studies included in the meta-analysis differed in terms of whether their alpha coefficient was above .80 (e.g., Brutton & Vanryckeghem, 2003; 2007) or not (Węsierska & Vanryckeghem, 2015). In some studies, alpha values were not reported (e.g., Beilby et al., 2012; Bernardini, Vanryckeghem, Brutton, Cocco, & Zmarich, 2009; Kawai, Healey, Nagasawa, & Vanryckeghem, 2012). Although missing or low alpha values may indicate methodological weaknesses in primary studies, Valentine (2009) did not recommend that meta-analyses should exclude studies based on low or missing alpha values. This because there may be other reasons for not reporting on alpha values than methodological weaknesses (e.g., journal requirements). However, as measurement instrument reliability is important information, the alpha coefficients reported in the studies should have been included in the characteristics of the primary studies (Table 1 in Paper 1).

As a new measurement tool was developed for Study 2, both inter-rater reliability and internal consistency were assessed. The investigation of inter-rater reliability was motivated by the widespread use of caregiver proxies in examining impacts on young children and by the lack of research investigating the reliability of caregiver reports. As measured by ICC, inter-rater reliability relates the degree of disagreement between raters to the differences between cases. Consequently, this can also reflect an instrument's ability to differentiate between participants as rated by several raters. As most of the ICCs indicated good inter-rater reliability, these results were interpreted to indicate good reliability of OASES-C.

5.4 Main limitations

The principal limitation of this PhD study concerns the number of participants included in Study 2. Jones, Gebski, Onslow and Packman (2002) pointed out that recruiting a large sample is often challenging because of the relatively low prevalence of stuttering. The features of variability and natural recovery are believed to make recruitment even more challenging; stuttering can be under-diagnosed in many cases because it can be difficult to differentiate from normal disfluency and/or because it is likely to resolve spontaneously. As the qualitative data indicated parental reluctance to talk with their children about stuttering and impact, the aim of the study may also have influenced recruitment. While the sample size was considered adequate for investigation of caregivers' perceptions of impact, the generalizability of results is limited. Furthermore, a larger sample would also have facilitated analysis of factors associated with impact of stuttering and psychometric investigation of the newly developed assessment instrument.

6. Ethical considerations

6.1 Ethical considerations before, during and after data collection

Research ethics refers to values, norms and institutional arrangements that contribute to constitution and regulation of scientific activity (De nasjonale forskningsetiske komiteene, 2009). The ethical principles in research include norms for the relationship between researchers, the considerations for the research subjects and the responsibility of the use and presentation of the data. An important standard for ethical considerations in research is that the research shall have a purpose and value expanding beyond itself and the researchers own circle (Backe-Hansen, 2012). In this PhD project, the aim for the planned studies was to investigate impact of stuttering in young children. Hopefully will the knowledge from this specific project contribute to increased knowledge in adults working or having responsibility for children who stutter about how stuttering can affect young children. If applied, the OASES-C may reveal impact and treatment needs in Norwegian children who stutter.

Data collection, anonymization and storage of data was planned and approved by Norwegian Center for Research Data before the project started (see appendix 1). Due to challenges with recruitment and changes in the questionnaires included, they were contacted several times for approval of changes (see appendix 2). After the data collection, the data was anonymized and safely stored in accordance with the guidelines by the Norwegian Center for Research Data. To secure full anonymity, no quotes or information that could lead to identification of children were included in presentation of the qualitative data.

For the children, participation involved playing with undersigned and a kindergarten teacher when speech was recorded and assessment with a battery of language tests. The children were given age-adjusted information about the study and what their participation involved by parents on beforehand and by undersigned when meeting the children. All children consented to participate before the assessment started. During assessment, the children were motivated to complete the assessment, but if a child seemed unwilling or unmotivated we either took a break or ended the session. With some individual adjustments all of the children completed the assessment. For one child for instance, the kindergarten teacher was present during the whole assessment due to shyness. For some children, not all of the language tests were completed.

6.2 Children in research

Special consideration needs to be taken when children are involved in research as children have particular needs for protection (NESH, 2016). Children in all ages can be affected by being asked to report on their feelings and thoughts. However, specific considerations need to be taken in regards to the young children how stutter, as they are in a vulnerable situation where the further stuttering pathway is not mapped out yet. In Study 2 it was decided to apply parent report in assessment of impact and not include children's self-report. Before discussing this decision it needs to be pointed out that ethical challenges and considerations may vary across studies and cultures. Variation is also found within the present PhD -project, as both self-report (Study 1) and parent-report (Study 2) were applied. Notably, differences in methodological choices do not have to mean differences in how ethical the different studies are, it rather reflects the different conditions the studies are carried out within.

In this particular study, several issues affected the decision of applying parent report. First, initial contact with the parents revealed reluctance to including the children. The qualitative data described in Paper 2 revealed the same: parents are reluctant to talking about stuttering and impact with their children. Second, the condition the self-report would be carried out within was not considered to attend to the children's needs appropriately as this would have involved asking the children to report on how stuttering affected them and then not provide any help for this. Third, as disorder-specific questionnaires such as the OASES contain items specifically targeting the negative effects of stuttering, some children may get an impression that it is expected that stuttering should have a negative effect. Potentially, this can affect their later feelings and thoughts related to stuttering. However, it needs to be pointed out that asking children how they are feeling, what they are thinking or what they have experienced can be done in a manner that attends to the children's needs and without exposing the child for any risk of any negative effects. Such situations can have positive consequences such as the child experiencing being seen or understood.

7. Discussion of the impact of stuttering in young children

7.1 How stuttering affects young children

Two factors are considered especially important in understanding the impact of stuttering. First of all, stuttering is a disorder that disrupts one of the most fundamental aspects of human life: communication. For this reason, stuttering has the potential to affect all kinds of relationships and interactions for the speaker. Secondly, as a group, children who stutter are known to be more negative in mood (Eggers et al., 2010; Ntourou et al., 2013) and more reactive, with poorer emotion regulation (Karrass et al., 2006) than children who do not stutter. Along with environmental influences, these temperamental characteristics can put these children at risk of stronger and more frequent reactions related to their stuttering, further exacerbating its frequency, duration and severity (Karrass et al., 2006). As more negative emotionality predicts persistency (Ambrose et al., 2015), these temperamental characteristics may also affect the stuttering pathway.

7.1.1 Main findings

The summary of self-report studies in Paper 1 indicates that stuttering in young children is associated with negative communication attitudes. Results in Papers 2 and 3 indicate that parents and kindergarten teachers perceive children to be affected by their stuttering—that is, stuttering is associated with the children's own reactions and their struggles in communication situations. The results also indicate that quality of life is affected to a little extent in terms of reduced self-confidence, negative mood and changes in peer interaction and peers' reactions. These results align with earlier self-report studies investigating the impact of stuttering in school-aged children (see Lankman et al., 2015), indicating that stuttering can have an adverse effect on young children's lives soon after stuttering onset.

Taken together, these findings are robust, as the four informant groups considered most informed in this context (children themselves, mothers, fathers and kindergarten teachers) all perceived stuttering as influencing the children's lives. Additionally, the caregivers showed good degree of agreement when reporting impact. All three studies found that impact of stuttering including communication attitudes are a common, but not inevitable part of the stuttering disorder among young children; results from the primary studies in Paper 1 indicated an absence of negative communication attitudes in some children, and some

children in Study 2 also experienced minimal impact, according to all three caregivers (see Table 2 in Paper 3). Nevertheless, as impact was revealed in the majority of children, either through self-report of communication attitudes or caregiver report of overall impact, these findings highlight the importance of assessing the impact of stuttering on young children.

The quantitative findings indicated strong associations between different aspects of health states in young children who stutter as framed by the ICF and operationalized in OASES-C, including children's reactions to stuttering (personal factors), communicative struggle (activity limitation) and quality of life (participation limitation). The qualitative findings provided further insights into these relationships; parents perceived activity limitation—especially how children are affected in communication situations—to be associated with listeners' attentiveness and reactions. Some parents also indicated that stuttering was to some extent associated with either activity limitation or participant restriction because of the children's personality (personal factors; e.g., outgoing, social). As described by the parents, some personality traits were considered to be protective against impact. Traits were not attributed to children who experienced more severe impact, although it seems likely that traits involving negative mood, low emotion regulation and high emotion activation as described in the temperament literature can render children prone to more severe impact. In this regard, Guitar (2014) stated that a person's experience of disability (activity limitation) and handicap (restrictions in participation) had more to do with how the person and significant others responded to stuttering than with the severity of the stuttering.

7.2 Multi-informant assessment informants

Overall, these results highlight the importance of assessing the various aspects of impact with different informants, including children reporting on present thoughts and feelings, caregivers on behavior, emotional expressions related to stuttering, communicative struggles across situations and quality of life. As kindergarten teachers observe these children's interactions with peers for many hours each day, it is especially important to assess from these caregivers' perceptions of communication difficulties with other children and reactions from others. Inclusion of the children themselves along with parents and kindergarten teachers seems to deliver a more reliable and comprehensive assessment of impact, as informants develop insight and can report on different aspects. This aligns with the

conclusion of Eiser and Morse (2001) that there are strong arguments for obtaining information from both parents and children when possible.

Agreement between parent and child reports varies across studies, depending on the instruments used and HRQoL domains investigated (see section 3.3.2). However, Eiser and Morse (2001) among others have stated that agreement is lower when the children's inner states are reported on. On a related point, results in Paper 2 indicated that parents' insight into impact varied across OASES-C sections; parents perceived themselves to have least insight into reactions to stuttering (see Table 4 in Paper 2) and into cognitive reactions in particular. Surprisingly, caregivers showed good inter-rater reliability on impact ratings of reactions. This may be explained by the method applied; caregivers can exhibit good inter-rater reliability in their observations of impact while being less certain whether their perceptions reflect the children's experiences. This may indicate the need for self-reports when investigating internal states, as for instance in the use of KiddyCAT.

While the combination of self-reporting with reports by caregivers would seem to provide the most reliable assessment, it may be challenging in some cases to include the children. For instance, children may not be able to report on impact, or their parents may not want them to (as indicated in the qualitative data in Paper 2). In the one study that specifically investigated parental and child reports of impact, Ntourou et al. (September 2017) found that that mothers' ratings correlated with their children's ($r = .29$) while the fathers did not. In the literature it has also been suggested that mothers may have more insight into their children's well-being than the fathers (see section 1.3 Paper 3 for a discussion). Even though results presented in Paper 3 indicated good agreement between mothers, fathers and kindergarten teachers, there were also cases where the raters had contradictory perceptions. Therefore, if only one rater is chosen, this choice needs to be considered carefully and differences between raters acknowledged.

7.3 Implications for research and practice

Evidence of the negative impact of stuttering in young children highlights some important issues. First, young children should not experience their lives as adversely affected by stuttering, which should therefore be treated before the children experience the disorder's negative impact. Alternatively, as suggested by Onslow (2017, September), it may be that in the future, SLPs will be able to facilitate children's development fluency before stuttering

occurs, so avoiding these experiences. Secondly, there is clearly a need to assess impact and to consider appropriate treatment, as environmental reactions and the children's own reactions towards the stuttering may interact with genes in predicting the children's stuttering pathways (Dworzynski et al., 2007). As discussed above, impact assessment involving multiple informants can provide more reliable insights. SLPs should keep in mind that caregivers' reports are not a substitute for children's self-reports, and that dependence on caregiver reports alone risks overlooking certain aspects of impact.

7.3.1 Treatment of impact

As discussed previously in this chapter, the choice of informants must be carefully considered in assessment of impact as informants can perceive impact differently. In line with results by Ntourou et al. (2017), differences between young children and parents must be anticipated in assessment of impact. In interpreting these differences, it is assumed that raters' information is complementary, and that information from all raters should be taken into account when considering or providing treatment. In Study 2, the qualitative data indicated that several parents were uncertain of how to handle impact, and treatment targeting impact may well be needed, as parents' strategies for handling impact may be important for the child's overall well-being and further development.

The importance of including impact when treating young children has been highlighted by several; for instance Guitar (2014) suggested more direct intervention to deal with children's negative feelings if they are present. And in the Palin Parent-Child Interaction therapy (Palin PCI (Kelman & Nicholas, 2008) components of the program include building self-confidence and dealing with emotions (Botterill & Kelman, 2010). Additionally, Boyle (2015) noted the value of seeking to increase self-esteem and strengthening family social support, as these factors are associated with improved QoL in people who stutter, regardless of stuttering severity. Notably, the qualitative data in Paper 2 indicate that some of the parents' strategies for handling stuttering and impact involved precisely this—providing the children with love and support to strengthen their self-confidence.

However, specific programs targeting impact in young children who stutter has not been found. In a population with both preschool and school-aged children (aged 4-14), Byrd, Hampton, McGill, and Gkalitsiou (2016) investigated the effect of an intensive therapy

program targeting feelings concerning ability to talk, perceptions of ability to establish friendships and influence of stuttering on QoL. Pre- and posttest assessment with the OASES indicated that the therapy had significant effects on communication attitudes and impact of stuttering on quality of among the children aged 7-14 years. For both this group and for the younger group of 4-6 year olds, parents reported increase in the children's perceptions of ability to make friends and decrease in impact of stuttering on QoL. Byrd et al. (2016) cautioned the long term effects of this therapy, however, these results are promising as they indicate positive effects of treatment of impact in children who stutter. Even though treatment programs treating the cause of the impact (i.e., the stuttering) has been shown to reduce impact (e.g., KiddyCAT pre and post measure; de Sonnevile-Koedoot et al., 2015), it can be argued that temperamental characteristics and the surrounding environment make some children more prone to negative impact than others, and that these children may need treatment that also specifically targets impact.

7.4 Further development of OASES-C

The comprehensiveness and theoretical framing of OASES-C provides a good foundation for revealing novel aspects of impact in young children. It seems likely that the instrument's comprehensiveness also increases the likelihood of detecting children affected by stuttering, which is especially important for clinicians in this area.

The OASES instruments for school-aged children, teens and adults have been thoroughly investigated, and the results indicate good overall validity and reliability. This first evaluation of OASES-C indicates good construct validity and good reliability (see Paper 2 and 3), is considered an important initial step in validating the questionnaire. In particular, the inclusion of qualitative data provided important information about parents' observations of impact and about their perceptions of the instrument's content and a foundation for future adaptation.

The investigation of inter-rater reliability in caregivers' ratings provided novel information about agreement about impact. However, the instruments' properties will require further investigation, especially as OASES-C not only targets a different age group but also differs from other OASES instruments in terms of method. The most important issues to be addressed before introducing OASES-C to clinical settings are as follows. First, there were several indicators of construct irrelevance in Section I. a) Section I was not associated with

the other sections. b) The frequency of IDK and *not relevant* responses was highest for Section I. c) Parents' qualitative responses indicated that some items were not relevant for their child (e.g., knowledge of treatment). Interestingly, several items such as knowledge of stuttering and stuttering treatment were those that had no clear association with the ICF. One possible explanation may relate to universality—that is, these issues may not be universal across health states in general or specifically across age groups who stutter.

Second, alpha values above .90 on Section III may indicate that some items are redundant (Tavakol & Dennick, 2011). Analyses of redundancy using inter-item correlation would indicate whether there are items that do not contribute additional information. For instance, it may be that young children's struggles in communication situations are less variable across persons and settings, leaving caregivers to observe and report on small nuances. Thus, if some of the situation-specific questions add no additional information, these should be omitted. Third, as large correlations (above .50; Cohen, 1988) were found between OASES-C sections (see Table 3 in Paper 2), a factor analysis should be conducted to investigate whether OASES-C assesses four different but associated constructs as indicated by the different sections, or whether the adapted version assesses a different set of constructs.

The creation of OASES-C facilitates further future evaluation of the impact of stuttering. For example, it will be important to know how soon after the onset of stuttering children experience negative impacts. It will also be useful to explore specific factors within individual children and in their environment that may contribute to the experience of negative impact. Future studies should also investigate the value of OASES-C in determining treatment needs and its sensitivity in measuring treatment progress.

7.5 Conclusion

This thesis provides an insight into impact of stuttering as perceived across different groups of informants; mothers, fathers, kindergarten teachers and the children themselves all reported that stuttering affects the children's lives. The strengths of this comprehensive, multi-informant assessment of impact is the finding that impact is perceived similarly across caregivers. However, as caregivers' agreement and parents certainty in their ratings varied across aspects of impact, these findings highlight the importance of including several informants, preferably by including reports by the children themselves. Given the potential negative impact of stuttering on quality of life even in young children, a comprehensive and specific measure like OASES-C is suitable for assessment purposes as well as during treatment in order to measure outcomes. Finally, as the present findings indicate that stuttering can have an adverse impact on several aspects of children's lives, it is important that clinicians, health care professionals, researchers and health funding authorities work to ensure that children who stutter receive professional support early on.

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Dissertational Papers

I

II

III

Paper I:

Guttormsen, L. S., Kefalianos, E., & Næss, K. A. B. (2015).

Communication attitudes in children who stutter: A meta-analytic review. *Journal of Fluency Disorders*, 46, 1–14.

Paper II:

Guttormsen, L. S., Yaruss, J.S., & Næss, K. A. B. (2018). Parents' perceptions of the overall impact of stuttering in young children.

Submitted to *American Journal of Speech and Language Pathology*

Paper III:

Guttormsen, L. S., Yaruss, J. S., & Næss, K. A. B. (2018).

Caregivers' perceptions of stuttering impact in young children: Inter-rater reliability of mothers', fathers' and kindergarten teachers' ratings. Submitted to *Journal of Communication Disorders* (April 2018)

APPENDIX

Linn Guttormsen
Institutt for spesialpedagogikk Universitetet i Oslo
Postboks 1140 Blindern
0318 OSLO

Vår dato: 05.12.2014

Vår ref: 40576 / 3 /

Deres dato:

Deres ref:

TILBAKEMELDING PÅ MELDING OM BEHANDLING AV PERSONOPPLYSNINGER

Vi viser til melding om behandling av personopplysninger, mottatt 03.11.2014. All nødvendig informasjon om prosjektet forelå i sin helhet 04.12.2014. Meldingen gjelder prosjektet:

<i>40576</i>	<i>Stamming i førskolealder</i>
<i>Behandlingsansvarlig</i>	<i>Universitetet i Oslo, ved institusjonens øverste leder</i>
<i>Daglig ansvarlig</i>	<i>Linn Guttormsen</i>

Personvernombudet har vurdert prosjektet, og finner at behandlingen av personopplysninger vil være regulert av § 7-27 i personopplysningsforskriften. Personvernombudet tilrår at prosjektet gjennomføres.

Personvernombudets tilråding forutsetter at prosjektet gjennomføres i tråd med opplysningene gitt i meldeskjemaet, korrespondanse med ombudet, ombudets kommentarer samt personopplysningsloven og helseregisterloven med forskrifter. Behandlingen av personopplysninger kan settes i gang.

Det gjøres oppmerksom på at det skal gis ny melding dersom behandlingen endres i forhold til de opplysninger som ligger til grunn for personvernombudets vurdering. Endringsmeldinger gis via et eget skjema, <http://www.nsd.uib.no/personvern/meldeplikt/skjema.html>. Det skal også gis melding etter tre år dersom prosjektet fortsatt pågår. Meldinger skal skje skriftlig til ombudet.

Personvernombudet har lagt ut opplysninger om prosjektet i en offentlig database, <http://pvo.nsd.no/prosjekt>.

Personvernombudet vil ved prosjektets avslutning, 01.03.2016, rette en henvendelse angående status for behandlingen av personopplysninger.

Vennlig hilsen

Removed due to privacy protection

Removed due to privacy protection

Kontaktperson: Removed due to privacy protection

Vedlegg: Prosjektvurdering

Dokumentet er elektronisk produsert og godkjent ved NSDs rutiner for elektronisk godkjenning.

Avdelingskontorer / District Offices:

OSLO: NSD, Universitetet i Oslo, Postboks 1055 Blindern, 0316 Oslo. Tel: +47-22 85 52 11. nsd@uio.no

TRONDHEIM: NSD, Norges teknisk-naturvitenskapelige universitet, 7491 Trondheim. Tel: +47-73 59 19 07. kyrre.svarva@svt.ntnu.no

TROMSØ: NSD, SVF, Universitetet i Tromsø, 9037 Tromsø. Tel: +47-77 64 43 36. nsdmaa@sv.uit.no



FORMÅL

Formålet med prosjektet er å kartlegge tidlig stammeutvikling og faktorer som er antatt å innvirke på stammeutviklingen. Både barn som stammer og barn som ikke stammer skal kartlegges, dette for å undersøke om det er eventuelle forskjeller mellom gruppene. Faktorene som antas å innvirke på stammeutviklingen er temperament, språkferdigheter og kommunikasjonsholdninger.

UTVALG

Utvalget består av barn i alderen 3-7 år som stammer og som ikke stammer, samt deres foreldre og logoped.

Barna som stammer rekrutteres ved at behandlende logoped gir informasjon om forskningsprosjektet. Barn som ikke stammer rekrutteres fra barnehager i østlandsområdet.

INFORMASJON OG SAMTYKKE

Utvalget informeres skriftlig om prosjektet og samtykker til deltakelse. Reviderte informasjonsskriv mottatt 04.12.2014 er godt utformet.

DATAMATERIALETS INNHOLD

Det behandles sensitive personopplysninger om helseforhold.

INFORMASJONSSIKKERHET

Personvernombudet legger til grunn at forsker etterfølger Universitetet i Oslo sine interne rutiner for datasikkerhet. Dersom personopplysninger skal sendes elektronisk, bør opplysningene krypteres tilstrekkelig.

Questback er databehandler for prosjektet. Universitetet i Oslo skal inngå skriftlig avtale med Questback om hvordan personopplysninger skal behandles, jf. personopplysningsloven § 15. For råd om hva databehandleravtalen bør inneholde, se Datatilsynets veileder: <http://www.datatilsynet.no/Sikkerhet-internkontroll/Databehandleravtale/>. Personvernombudet ber om kopi av avtalen for arkivering (sendes: personvernombudet@nsd.uib.no).

PROSJEKTSLUTT OG ANONYMISERING

Forventet prosjektslutt er 01.03.2016. Ifølge prosjektmeldingen skal innsamlede opplysninger da anonymiseres. Anonymisering innebærer å bearbeide datamaterialet slik at ingen enkeltpersoner kan gjenkjennes. Det gjøres ved å:

- slette direkte personopplysninger (som navn/koblingsnøkkel)
- slette/omskrive indirekte personopplysninger (identifiserende sammenstilling av bakgrunnsopplysninger som f.eks. bosted/arbeidssted, alder og kjønn)
- slette lydopptak

Vi gjør oppmerksom på at også databehandler (Questback) må slette personopplysninger tilknyttet prosjektet i sine systemer. Dette inkluderer eventuelle logger og koblinger mellom IP-/epostadresser og besvarelser.

Til foreldre/foresatte til
barn som stammer

Oslo 15.03.16

Forespørsel deltakelse i forskningsprosjektet ”Språkferdigheter, kommunikasjons holdninger og temperament hos barn som stammer og barn som ikke stammer”

Det overordnede formålet med denne studien er å få mer kunnskap om tidlig stammeutvikling. Dette gjøres ved å kartlegge faktorer som antas å innvirke på stammeutviklingen hos barn som stammer og barn som ikke stammer. Kunnskap om tidlig stammeutvikling vil kunne gi en bedre forståelse av stamming og føre til en mer målrettet oppfølging slik at flere barn slutter å stamme. Studien ønsker derfor å følge utvikling på noen sentrale områder hos barn som stammer og barn som ikke stammer for å kartlegge eventuelle forskjeller mellom gruppene og innad i gruppen med barn som stammer. Studien vil ha en varighet på 12 måneder og er en del av et doktorgradsprosjekt (2013-2017) som gjennomføres ved Institutt for spesialpedagogikk.

Hva innebærer deltakelse i studien?

Deltakelse i studien vil innebære at enten barnets logoped eller prosjektleder/prosjektmedarbeidere tar opptak av barnets tale når barnet leker, vurderer grad av stamming på bakgrunn av taleopptaket og kartlegger barnets språkferdigheter (varighet ca 60 minutter).

For foreldrene innebærer deltakelse i studien utfylling av 3 spørreskjemaer vedrørende barnets reaksjoner på stammingen, barnets temperament og hvordan stammingen har påvirket barnet ved oppstart og avslutning av studien (varighet ca 30 minutter). Foreldrene vil bli intervjuet over telefon ved oppstart av studien av prosjektleder/prosjektmedarbeidere. I tillegg bes førskolelærer om å besvare to av spørreskjemaene som foreldrene har besvart vedrørende barnets reaksjoner på stammingen og temperament (varighet 20 minutter). Både foreldre og eventuell logoped som jobber med barnet vil bli spurt om å vurdere grad av stamming månedlig (varighet 1-2 minutter).



Et samtykke til å delta i studien vil innebære at både foreldre og barn deltar i studien. Dersom du/dere ønsker innsyn i spørreskjemaene og testene som brukes med barna, kan en slik forespørsel sendes til prosjektleder. Prosjektleder vil gå igjennom testene dersom det er ønskelig, og kan også gi en kort rapport fra språkkartleggingen av barnet.

Alle personopplysninger vil bli behandlet konfidensielt. Hvert barn blir tildelt et løpenummer og navnelisten med løpenumrene blir oppbevart i et låst skap under prosjektperioden. Resultatene av studien vil bli publisert som på gruppenivå, uten at det enkelte barn kan gjenkjennes. Studien avsluttes innen 15.04.2018. Innsamlede data vil være anonymisert før studien avsluttes. Prosjektet er tilrådd av Personvernombudet for forskning, Norsk samfunnsvitenskapelig datatjeneste (NSD).

Det er helt frivillig å delta i studien, og du/dere kan når som helst trekke ditt/deres samtykke uten å oppgi noen grunn. Hvorvidt du/dere ikke vil delta i studien eller ønsker å trekke dere fra den, har ingen betydning for logopedtilbudet barnet får eller barnets situasjon i barnehagen.

Dersom du/dere samtykker til at ditt/deres barn kan delta i studien, vennligst signer den vedlagte samtykkeerklæringen og returner den til barnehagen så snart som mulig. Samtykkeerklæringen blir sendt til prosjektleder.

Har du spørsmål i forbindelse med denne forespørselen, vennligst kontakt undertegnede.

Med vennlig hilsen,

Linn Stokke Guttormsen

Stipendiat ved Institutt for spesialpedagogikk

e-post: l.s.guttormsen@isp.uio.no

Samtykkeerklæring

Jeg/vi har mottatt skriftlig informasjon om forskningsprosjektet «*Språkferdigheter, kommunikasjonsholdninger og temperament hos barn som stammer og barn som ikke stammer*».

Jeg/vi gir samtykker til at (navnet på barnet ditt) deltar i studien.

Signatur:

Navn i blokkbokstaver:

E-postadresse:

Telefonnummer:

Linn Stokke Guttormsen

Fra: Removed due to privacy protection
Sendt: 21. desember 2015 15:02
Til: Linn Stokke Guttormsen
Emne: Prosjektnr: 40576. Stamming i førskolealder

BEKREFTELSE PÅ ENDRING

Hei, viser til endringsmelding kommet inn som epost registrert 26.11.2015.

Vi har nå registrert følgende endringer:

- spørreskjema til foreldre og barnehage utvides med noen spørsmål om hvordan barna reagerer på stammingen
- dato for prosjektslutt utsettes til 1. mars 2017
- UiO sitt nettskjema skal brukes som databehandler og ikke Questback

Personvernombudet forutsetter at prosjektopplegget for øvrig gjennomføres i tråd med det som tidligere er innmeldt, og personvernombudets tilbakemeldinger. Vi vil ta ny kontakt ved prosjektslutt.

--

Med vennlig hilsen

Removed due to privacy protection

Rådgiver NSD Personvern

Tlf. direkte: Removed due to privacy protection

Tlf. sentral: 55 58 81 80

Internettadresse: www.nsd.uib.no/personvern

Linn Stokke Guttormsen

Fra: Removed due to privacy protection
Sendt: 30. mars 2017 14:52
Til: Linn Stokke Guttormsen
Emne: Prosjektnr: 40576. Stamming i førskolealder

BEKREFTELSE PÅ ENDRING

Hei, viser til epost mottatt 28.3.17.

Vi har nå registrert at dato for prosjektslutt og anonymisering endres fra 1.3.17 til 30.12.17. Utvalget informeres.

Personvernombudet forutsetter at prosjektopplegget for øvrig gjennomføres i tråd med det som tidligere er innmeldt, og personvernombudets tilbakemeldinger. Vi vil ta ny kontakt ved prosjektslutt.

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Med vennlig hilsen

Removed due to privacy protection

Seniorrådgiver | Senior Adviser
Seksjon for personverntjenester | Data Protection Services
T: (+47) ^{Removed due to privacy protection}

NSD – Norsk senter for forskningsdata AS | NSD – Norwegian Centre for Research Data Harald Hårfagres gate 29,
NO-5007 Bergen
T: (+47) 55 58 21 17
postmottak@nsd.no www.nsd.no

Linn Stokke Guttormsen

Fra: Removed due to privacy protection
Sendt: 22. januar 2018 13:11
Til: Linn Stokke Guttormsen
Emne: Prosjektnr: 40576. Stamming i førskolealder

BEKREFTELSE PÅ ENDRING

Hei, viser til endringsmelding registrert hos personvernombudet 22.01.2018.

Vi har nå registrert at dato for prosjektslutt og anonymisering forlenges fra 30.12.2017 til 15.04.2018. Vi har videre registrert at det samles inn data om flere barn, på samme måte som tidligere.

Personvernombudet forutsetter at prosjektopplegget for øvrig gjennomføres i tråd med det som tidligere er innmeldt, og personvernombudets tilbakemeldinger. Vi vil ta ny kontakt ved prosjektslutt.

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Med vennlig hilsen

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Seniorrådgiver | Senior Adviser
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