# Lexical development in children with Down syndrome:

## A communicative perspective

Stijn R. J. M. Deckers

Behavioural
Science
Institute

# Lexical development in children with Down syndrome: A communicative perspective

Stijn R. J. M. Deckers

The research presented in this dissertation was supported by a grant from Nationaal Regieorgaan Praktijkgericht Onderzoek SIA (RAAK PRO 3-39) awarded to dr. Yvonne van Zaalen and was carried out at the School for Allied Health Professions of the Fontys University of Applied Sciences and at the Behavioural Science Institute of the Radboud University, the Netherlands.

**ISBN**
978-90-826784-0-6
**Cover photography**
Kim's Fotografie & Creatie
**Cover models**
Pien and Gijs Bex
**Design/lay-out**
proefschrift-aio.nl
**Print**
proefschrift-aio.nl

# Lexical development in children with Down syndrome: A communicative perspective

## Proefschrift

Ter verkrijging van de graad van doctor
aan de Radboud Universiteit Nijmegen
op gezag van de rector magnificus prof. Dr. J.H.J.M. van Krieken,
volgens besluit van het college van decanen
in het openbaar te verdedigen op donderdag 29 juni 2017
om 12.30 uur precies

door

**Stijn Robertus Jakob Maria Deckers**
geboren op 31 augustus 1987
te Heerlen

**Promotoren**
Prof. dr. L. T. W. Verhoeven
Prof. dr. H. van Balkom

**Copromotor**
Dr. Y. van Zaalen (Fontys Hogescholen)

**Manuscriptcommissie**
Prof. dr. R. Didden
Prof. dr. B. Maassen (Rijksuniversiteit Groningen)
Prof. dr. I. Zink (Katholieke Universiteit Leuven, België)

*"All speech, written or spoken, is a dead language,*
*Until it finds a willing and prepared hearer"*

Robert Louis Stevenson
Reflections and Remarks on Human Life

# Contents

# Chapter 1

## General introduction

# General introduction



King Willem-Alexander of the Netherlands stated in 2013 that the "participation society" is emerging, as a shift from the "welfare society". Participation has become the key concept in policy making and is extensively used, both in clinical practice and in research, also since the World Health Organization introduced its definition in the International Classification of Functioning in 2001. Social participation are those activities undertaken inside and outside the house, allowing individuals to meet others, contribute to society, and to stay involved in that society (Broese van Groenou & Deeg, 2006). To reach optimal levels of participation, our highly linguistic, human society asks for well-developed language and communication skills. Young children with Down syndrome often experience difficulties in both their language and communication development interfering with their opportunities for participation. Difficulties include a late onset of the first spoken word, also related to a high degree of speech unintelligibility, and a delay in sequential lexical development. In typically developing children, word learning emerges naturally from situations in which children are engaged in social interactions. Accordingly, lexical development should be studied in light of participation and engagement in communicative contexts. Therefore, the present thesis will focus on the lexical development of young children with Down syndrome in a communicative perspective.

This introductory chapter starts out with a description of the Down syndrome behavioural phenotype. After a description of the communicative perspective on lexical development and an overview about the lexical development of children with Down syndrome, the two main themes of this thesis are introduced: measuring lexical development and predictors of lexical development in children with Down syndrome. In a final section, the research questions of the present study will be presented along with an outline of this thesis.

## The Down syndrome behavioural phenotype

Research has shown that many children with intellectual disabilities experience difficulties in language and communication development (Kaiser, Hester, & McDuffie, 2001; Rondal, 2001). Several studies in intellectual disabilities did, however, not find the level of intellectual disability to be predictive of later language skills (e.g. Calandrella & Wilcox, 2000; McCathren, Yoder, & Warren, 1999). Individual differences in language development of children with intellectual disabilities are therefore largely ascribed to syndrome differences (Rondal, 2001). Vandereet (2010) states that various genetic syndromes manifest distinct phenotypes with unique communication and language characteristics.

She therefore stressed the need for future research to explore etiology-specific language phenotypes. For the Netherlands, the prevalence of intellectual disabilities is around 7 per 1000 live births (Wullink, Van Schrojenstein Lantman-de Valk, Dinant, & Metsemakers, 2007). The most common genetic cause of intellectual disability is Down syndrome (Sherman, Allen, Bean, & Freeman, 2007). Birth prevalence for Down syndrome in the Netherlands in 2003 was estimated at 14 per 10.000 live births and in 2010 at 7.7 per 10.000 live births (De Graaf et al., 2011). These numbers indicate that of all live births of children with intellectual disabilities in the Netherlands, around 11 – 20 % is diagnosed with Down syndrome. Given this relatively high prevalence rate, Down syndrome is the most valid reference group to further explore etiology-specific language phenotypes.

### Down syndrome in the Netherlands

Between 2008 and 2009, the Dutch Down Syndrome Foundation conducted a large questionnaire study amongst parents of children and adolescents with Down syndrome (De Graaf, 2012). Results indicated that between 52 - 58% of the sample were male, showing an overrepresentation of boys in Down syndrome diagnoses. In the age range of 0-5 years, about 17% of the children were from non-Western backgrounds, for the age group of 5-13 year-olds, these percentages were 15 and 5, respectively. Educational background of parents of children with Down syndrome was estimated low in 17% of the families, 45% as moderate, and 38% was estimated as high. De Graaf (2012) also asked about care professionals associated with the health care of the children and adolescents with Down syndrome. Of the 0-5 year-olds, 91.8% visited a physiotherapist, and 82.9% visited a speech-language pathologist. Of the 5-13 year-olds, a decrease is seen in visits to the physiotherapist (80.8% is visiting), where an increase is seen in visits to the speech-language pathologist (i.e., 97.6%).

As in many other countries, since the 1980s, more and more children with Down syndrome are entering regular schools in the Netherlands. In the Netherlands most children with Down syndrome start their school career slightly later than other children, often between 4.5 and 5 years of age instead of immediately after their fourth birthday (De Graaf, Van Hove, & Haveman, 2013).

On basis of information of the Dutch Ministry of Education and of the Dutch Down Syndrome Foundation, De Graaf (2007) estimated that of all Dutch children with Down syndrome born since the 1990s, 56% started their school career in a regular school. Of the children with Down syndrome starting in regular education approximately 40% is still in a regular school at the end of primary education (at the age of 12 years). However, more recent numbers indicate that only 26% of children with Down syndrome is still in a regular school at age 11 (De Graaf et al., 2013).

Down syndrome is usually (i.e., in 95% of the cases) caused by the presence of an extra chromosome at location 21 (Trisomy 21) (Sherman et al., 2007) either in all cells, or, in the case of mosaicism, in only some cells. According to Dykens (1995), a behavioural phenotype implies that persons affected by a particular genetic syndrome are more likely to show specific features and abnormal behaviours. Down syndrome literature often mentions a Down syndrome behavioural phenotype. Cognitively, most children with Down syndrome fall into the mild to severe range for an intellectual disability, with a mean intelligence quotient (IQ) of about 50 (Chapman & Hesketh, 2000). The Down syndrome behavioural phenotype includes relative strengths in some aspects of visuospatial processing, such as visual memory and visual imitation, nonverbal communication and social functioning. The behavioural phenotype also includes relative deficits in verbal processing, verbal working memory, spatial memory and aspects of motor functioning, such as motor planning, hypotonia and hyperflexibility. Hyperactivity, aggression, stubbornness, disobedience, inattention and impulsivity have also been often documented (for a complete overview of the Down syndrome behavioural phenotype, see Fidler, 2005; Fidler, Most, & Philofsky, 2008).

Besides these general characteristics, Kumin (2008) also summed up the strengths and weaknesses in the communication profile of children with Down syndrome. The two most common relative strengths mentioned are semantics and pragmatics, while weaknesses of children with DS tend to include: (a) shorter phrases in speech, (b) short conversations with many communication breakdowns, (c) reduced speech intelligibility, (d) significantly delayed onset of and difficulties in expressive language, and (e) language skills below cognitive level, but receptive language skills above expressive language skills. Intelligibility (i.e., the extent to which a listener can receive the message intended by a sender) of speech is a particular issue in Down syndrome (Price & Kent, 2008; Rondal & Edwards, 1997). Kumin (1994) performed a survey of 937 parents of children

with DS and found that over 58% of parents reported that their children frequently had difficulty being understood by people outside of their immediate circle, with a further 37% having difficulty being understood at least some of the time. All of these features may affect the ability to create the precise articulations required for clear speech (Cleland et al., 2010). People with Down syndrome present with a specific physiological and anatomical profile, including a smaller than average oral cavity, hypotonia of muscles around the mouth, fusion of lip muscles, and extra lip musculature (Miller, Leddy, & Leavitt, 1999; Spender et al., 1995), and with a poor speech motor control (Kumin, 1994), all adhering to the reported intelligibility problems.

Language has been often described as the major area of deficit in Down syndrome, with most particular difficulties manifested in expressive language (Fowler, 1990). The language difficulties are described as specific part of the Down syndrome behavioural phenotype, adhering to the high degree of perceived communication problems in children with Down syndrome.

## Lexical development: A communicative perspective

Communication can be defined as an information exchange process between a sender and receiver in which the response from the receiver suits the intentions of the sender (Levelt, 1989, 1999, 2004). Communication skills are fundamental to participation in all aspects of life, such as family, friends, school, work, and community. These skills provide the power to express needs and wants, to share information, to develop social relationships, and to participate within expected social etiquette routines (Light, 1989). As Chomsky (1959) already stated, communication requires productivity. Human communication is multimodal in nature as people mostly use a combination of speech and non-speech modes to communicate. This association between verbal and non-verbal communication modes is highly influenced by the communicative context (Alant, Bornman, & Lloyd, 2006).

Typically developing children understand the communicative intentions of communication partners, including those expressed in linguistic utterances, around 9-12 months of age (Tomasello, 2003). Children's prelinguistic communication and cognitive skills, that develop from 6 to 18 months of age, lay the foundation for the emergence of first words during the second year of life (Dromi, 1999). Intentional communication, expressed by gestures and vocalizations, is usually evident between 9 and 12 months and increases in frequency over the second year of life. These behaviours allow toddlers to engage in turn-taking and object-focused conversations with communication partners before acquiring language (Lederberg & Beal-Alvarez, 2010). Gestures serve as a bridge to linguistic

communication. The use of representational gestures predicts early lexical development in typically developing toddlers (Zaidman-Zait & Dromi, 2007). The first words become part of the communication repertoire between 10 and 13 months (Fenson et al., 1993). Most 1-year-olds produce their gestural and linguistic utterances to get an adult to do something with respect to an object or event or to get an adult to share attention with them to that object or event (Bates, Camaioni, & Volterra, 1975).

> "Human linguistic communication is symbolic. Linguistic symbols are social conventions by means of which one individual attempts to share attention with another individual by directing the other's attentional or mental state to something in the outside word. (…) It is this mental dimension that gives linguistic symbols their unparalleled communicative power, enabling them to be used to refer to and to predicate all kinds of diverse perspectives on objects, events, and situations in the world." (Tomasello, 2003, p. 8).

This acquisition of language is the result of a process of social interaction involving shared activities (Stoel-Gammon, 2001). The social-pragmatic approach of word learning emphasizes the inherently social nature of language and word learning (Akhtar & Tomasello, 1998). In this approach, word learning emerges naturally from situations in which children are engaged in social interactions in which they are attempting to understand and interpret adult communicative intentions as expressed in utterances (Tomasello, 2003). Language learning is intrinsically social since words are learned in and for conversations (Bloom, 2000) with others and for the purpose of communicating with others (Akhtar & Tomasello, 2000). "Language learning takes place through communication and is propelled by communicative needs" (Kühn & Langner, 2012, p. 182), which makes language a communication phenomenon (Reichling, 1967). The capacity to provide information about experiences and events is critically important for children's social connectedness, communicative competence, constructive listening, cognitive processing, and classroom success (Proctor & Zangari, 2009).

From a variety of studies (e.g., Hagoort, 2007; Leppänen, Lyytinen, Choudhury, & Benasich, 2004; Mildner, 2008) it can be learned that the language system is not localised in one specific area of the brain, but develops into a complex neural network in the first years of life. This neuronal language network assembles "a new machine created out of various neurocognitive and social components that evolved initially in the service of completely different functions" (Tomasello & Slobin, 2005, p. xxvii). Language does not develop without social interaction and communicative encounters. The human urge to interact with our environment,

explains why, in many cases, the neuronal language network is capable of finding alternative paths in case of obstructions (Van Balkom, Deckers, & Stoep, 2017). This enables multimodal representations of language, such as gestures, manual signs, speech, tactile symbols (Jackendoff, 2002). Within a socio-neurocognitive approach, language learning is strongly interrelated with executive control (Anderson, Anderson, Jacobs, & Smith, 2008; Goldberg, 2009) and attention regulation (Posner & Rothbart, 2007; Posner & Fran, 2008), perception (Bahrick & Lickliter, 2002; Sato, Cavé, Ménard & Brasseur, 2010), working memory and short term memory (Baddeley, 2012; Lum, Conti-Ramsden Page & Ullman, 2012), cognitive functions (Anderson, Northam, Hendy & Wrennall, 2001; Mildner, 2008), orientation, adaptation, social-emotional state (Sperber & Wilson, 1995; Kaczmarek, 2002); self-care and self-help skills (Ylvisaker & Feeney, 2008), and motor skills and mobility (Mashal, Andric, Small, 2012; Cogan,Thesen, Carlson, Doyle, Devinsky & Pesaran, 2014). These domains are also recognized in several clinical neuropsychological assessment procedures and test batteries, as the NEPSY II (Korkman, Kirk & Kemp, 2010), LNNB-Luria-Nebraska Neuropsychological Battery (Golden, 2004) and HRNB-Halstead-Reitan Neuropsychological Battery (Broshek & Barth, 2000).

Lexical development (i.e. word learning and vocabulary development) is a fundamental building block in the acquisition of language, and is often identified as one of the 'special' components of language (Bloom, 2000; Hauser, Chomsky, & Fitch, 2002; Pinker & Jackendoff, 2005). Vocabulary, which encompasses all the words that are known (i.e., receptive vocabulary) and used (i.e., expressive vocabulary) by an individual, is an essential part of a language. Without sufficient levels of vocabulary knowledge, communication would suffer, since conveying a message would need at least a basic knowledge of words (Agdam & Sadeghi, 2014). Given the social nature of language development, the study of lexical development needs a communicative perspective as proposed by Van Balkom (2009). This communicative perspective is based on abovementioned core developmental domains associated with communication and language competence (see Figure 1). In this adaptation of Fröhlich and Haupt's (2004) model of the holistic development of language and lexical skills, lexical development is interrelated with these developmental domains in a communicative context. This interrelatedness infers that (a) when a developmental domain is impaired, this will affect the lexical development of an individual, and (b) better or well-developed developmental domains may compensate for the impairment in others, resulting in better lexical outcomes than might have been expected given (a).

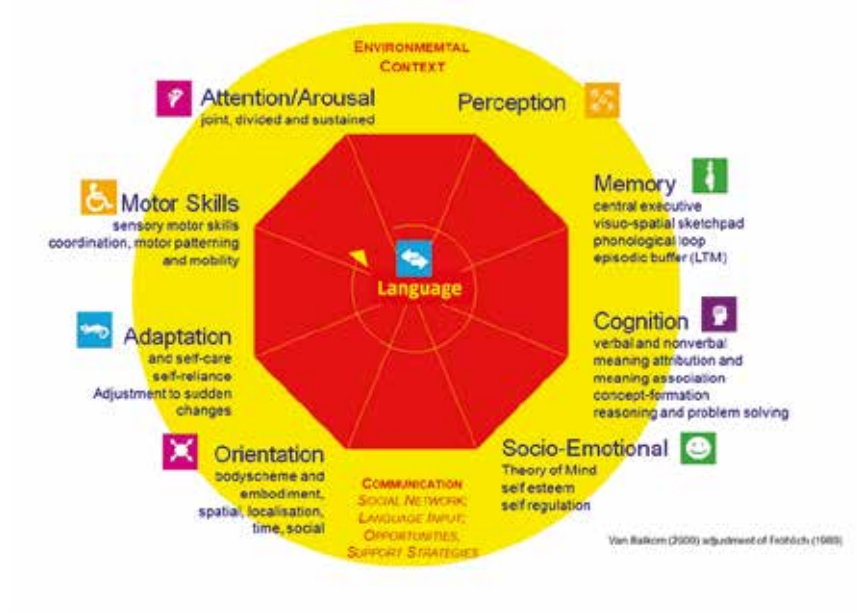



Figure 1. Model of holistic development of language and lexical skills (Van Balkom, 2009, adjustment of Fröhlich & Haupt, 2004).

## Lexical development in Down syndrome

The Down syndrome behavioural phenotype includes pronounced impairments in expressive language when compared to receptive language, including great deficits in expressive vocabulary size relative to mental age (Zampini & D'Odorico, 2013). The development of receptive vocabulary has often been found to be mental age appropriate in childhood (Abbeduto et al., 2003). Children with Down syndrome produce their first spoken word around the average age of 1;9 years [years;months] (Stoel-Gammon, 2001), though substantial individual variation exists (Berglund, Eriksson & Johansson, 2001). Contrary, Gillham (1990) found that it is not until the age of 3;9 years that some children uttered their first spoken words. Once they begin to produce words, their progress and development in lexical size relative to their typically developing peers is very slow (Brady et al., 2004; Yoder & Warren, 2004). Berglund et al. (2001) found the 50-word milestone was reached by about 25% of the children at age three, by about 50% at age four and about 75% at age five, where typically developing children reach this milestone around 18 months. About 10 - 20% of the children with Down syndrome produced less than 10 words in their spoken vocabularies

when they were already 3;0 – 5;0 years old. Some children had not even started to talk at all at age 5. The lexical size of children with Down syndrome at ages 3;0 and 4;0 in the Berglund et al.-study was comparable with that of typically developing children at ages 1;4 and 1;8 respectively. However, as in most studies of lexical development in Down syndrome (Galeote, Soto, Checa, Gómez, & Lamela, 2008), they only focused on spoken words as a measure of expressive vocabulary. The study of lexical development in children with Down syndrome often presents an important limitation, in that vocabulary production is taken into account only in its spoken modality, whereas many children with Down syndrome spontaneously produce several gestures and manual signs (Galeote et al., 2008). Miller et al. (1991, cited in Chapman, 1995) found that when also signed words are taken into account, the obtained vocabulary scores might be similar to mental age matched peers, indicating that the often found gap between expressive vocabulary and mental age may be highly due to assessment bias. Between the ages of 1;0 - 5;6 years 87% of 330 DS children used speech and manual signs simultaneously (Berglund et al., 2001). Given this high percentage of manual sign use, most studies in Down syndrome populations may have underestimated their lexical development by excluding signs. In order to gain more insight in the lexical development of children with Down syndrome, the number of signs or words produced via other communication means as graphic symbols, should be included in the measurement of vocabulary size.

## Measuring lexical development

Lexical development is typically measured by estimating the number of produced words by children at a particular age (Lederberg & Beal-Alvarez, 2010). The expressive vocabulary size of children with Down syndrome in clinical practice is often measured in one or multiple ways: standardized tests, spontaneous language analyses and/or parental reports. All have their own advantages and disadvantages. Standardized tests in clinical or laboratory settings have the advantage of being based on observable behaviour at a specific moment in time, which can be related to normative data. However, the attention span of young children is an important factor influencing the assessment and therefore the result is likely to prove unrepresentative of the child's abilities (Feldman et al., 2005). Secondly, standardized tests often require the child to interact with an unfamiliar adult, possibly influencing the child's responses (Pan, Rowe, Spier, & Tamis-Lemonda, 2004), which may be particularly true for children with DS (Miller, Sedey, & Miolo, 1995).

In standardized tests, language is provoked in an isolated situation and may not represent active use of these words for a specific child during the day. Words are provoked within a constraint time frame, conflicting with the fact that children with DS are known to have a slower response time (Inui, Yamanishi, & Tada, 1995). Spontaneous language analysis has the advantage to provide insight into how the child actually uses language in interaction with communicative partners, and are thus potentially more ecologically valid (Pan et al., 2004). However, language samples recorded in conversation with a researcher or a parent may be highly influenced by personality and contextual factors, such as person, setting, and materials used or included (Feldman et al., 2005; Yont, Snow, & Vernon-Feagans, 2003). Recordings of parent-child interactions within the clinic may consistently lead to underestimation of vocabulary size, because (a) young children talk less when they are in unfamiliar settings or around people they do not know, and (b) it is impossible to simulate the wide variety of situations in which a child produces language, and much of this language is context-bound (Mervis & Becerra, 2003). All these factors are less likely to influence assessment via parental reports. Researchers have assessed early lexical development by asking parents to report their children's vocabulary on word checklists, of which the MacArthur-Bates Communicative Development Inventories (CDI; Fenson et al., 1993) is also the most commonly used in clinical practice. The specific words in most early reported lexicons (i.e. lexicon size < 35 words) are remarkably similar when you compare the CDIs of typically developing children, deaf children (Lederberg & Beal-Alvarez, 2010) as well as of young children with Down Syndrome (Te Kaat-Van Os, 2013).

## Parental reports of lexical development: Communicative Development Inventories

When characteristics of clinical populations differ from those of typical populations in important ways, this affect the validity of test instruments, such as the use of the CDI in Down syndrome (Galeote, Checa, Sánchez-Palacios, Sebastián, & Soto, 2016). The validity of parental reports may be compromised in children with developmental disabilities, since parents may over- or underestimate their child's performance (Miller et al., 1995). This underlines the importance of assessing the validity of the CDI for use with children with Down syndrome. Galeote et al. (2016) studied the concurrent validity and reliability of the CDI for specific use in children with Down syndrome. Galeote et al. (2016) modified the CDI to also assess spontaneously produced gestures representing only concrete lexical items and found the concurrent validity and reliability to be high. In Dutch clinical practice, as a part of an Augmentative and Alternative Communication (AAC)

intervention, the use of Nederlands Ondersteund met Gebaren [Sign Supported Dutch] is a common intervention strategy in young children with DS, as a result of which these children use signs as an important mode of communication. Up to 5 years of age, manual signing may be the primary communication mode for many (Dutch) children with Down syndrome (Kumin, 2003). The number of used signs should thus be accounted for when assessing the expressive vocabulary development of children with Down syndrome and the CDI needs a modification. However, the validity of such a modification is unclear.

For the Dutch field, the N-CDI (N stands for 'Nederlands' ['of the Netherlands']) was earlier modified to include the modality of the words produced (Vandereet, Maes, Lembrechts, & Zink, 2011). Next to speech, parents could also indicate which words were expressed via a manual sign by their child. Although not the primary objective of Vandereet and colleagues, the concurrent validity with spontaneous utterances of 23 children with intellectual disabilities aged 3 to 6 years was investigated. Vandereet et al. (2011) concluded that their results provided preliminary support for the concurrent validity of the N-CDI as a measure of modality of expressive vocabulary. Although the N-CDI is the most often used assessment method of lexical development in children with Down syndrome, both in clinical practice and in research, the validity of these parental reports are still unclear.

## Spontaneous language analysis: Assessment of functional word use

Although the CDI represents a complementary approach that provides a complete and representative idea of the language and communicative abilities of children with Down syndrome (Galeote et al., 2016), a recognized limitation of the CDI is that it does not provide information about word frequency and functional word use (Galeote et al., 2008). Using the N-CDI provides us with insight in the different words that a child uses, and thus in the total number of words in a child's lexicon, but not in how they actually use these words in everyday settings. An interesting quantitative issue regarding the production of language is which words children use most often in spontaneous language (Tomasello, 2003). When looking at results based on CDIs, children with Down syndrome often show a deficit in the use of function words when compared to content words (Galeote et al., 2008), which may be heavily influenced by the overrepresentation of nouns and verbs in the CDI when compared to the number of function words. Another explanation may be that, in children with Down syndrome, the transitioning from 1- to 2-word speech is generally delayed (Chapman, Hesketh, & Kistler, 2001; Iverson et al., 2003). After this transitioning, children with Down syndrome have

a shorter mean length of utterance (MLU) than nonverbal mental age matched typically developing children (Chapman, Seung, Schwartz, & Kay-Raining Bird, 1998). Also, difficulties in the use of grammatical function words and tense and non-tense bound morphemes (Eadie, Fey, Douglas, & Parsons, 2002), and in the number of grammatical, and lexical verbs per utterance (Hesketh & Chapman, 1998) can be seen in comparison with MLU-matched controls.

In early development, children learn more nouns than other types of words (Tomasello, 2003). Gentner (1982) referred to this as the Natural Partitions hypothesis: nouns that children learn early in their development are prototypically used to refer to concrete objects, since concrete objects, that are spatially bounded, are more easily individuated from their environments than are mental or emotional states, actions, processes and attributes. Nouns and other open-class, content words show relative cognitive dominance, whereas relational words (i.e., function words) mostly serve to provide linguistic connections among the referential content words (Gentner & Boroditsky, 2001). Brent and Siskind (2001) showed that up to 60% of children's first words are those words that their parent has used relatively often as single-word utterances, such as names, interjections, performatives, and some object and action words.

The opportunities to learn language are closely linked to the Cognitive Linguistic notion of 'entrenchment' (Zenner, Speelman, & Geeraerts, 2014). Entrenchment refers to the degree of automatization of a cognitive unit resulting from repeated exposure (Schmid, 2007). Zenner et al. (2014) distinguish two types of entrenchment. The more an individual uses a certain word and talks about a given concept, or experiences the use of this word by communication partners, the more entrenched the word becomes in the individual's cognitive apparatus (i.e., communicative entrenchment). Concepts can also be activated perceptually. Then, entrenchment results from frequent exposure to a concept in a child's daily activities (i.e., experiential entrenchment). According to Zenner et al., there is often a non-coincidence between experiential frequency of concept exposure and the communicative frequency of concept use. This is referred to as the 'toothbrush problem'. Toothbrushes are used every day, therefore a child would have multiple experiences with toothbrushes. However, the frequency of use of the word toothbrush in daily conversations is relatively low compared to other words. This is the case for many content words, that are highly situation- or person-specific.

Tomasello (2003) stated that "as soon as just a small amount of grammar begins, young children's utterances are peppered with a relatively small number of high-frequency lexical items such as certain pronouns and function words with highly recurrent discourse functions – with the more well-known nouns and

verbs, which are typically thought of as the prototypical items in young children's vocabularies, used relatively infrequently as their specific referents occur in the child's experience at only irregular intervals" (p. 81). The CDI does not provide any insight in how and how frequently words are used by children with Down syndrome in daily communicative settings. In other words, it provides no information about words that are more communicatively entrenched. This information can only be obtained by observing the spontaneous language use of children with Down syndrome in multiple contexts and with multiple communication partners. Words that are frequently used throughout several modes of communication and that can maximize the potential for spontaneous language generation, both in spoken and/or signed modalities, are called 'core vocabulary'. These core words provide the opportunity to be engaged in communication and interaction in a proper, efficient and relatively quick manner (Hill, Kovacs, & Shin, 2015; Weighton & Dodd, 2011). To date, only little is known about this functional word use of children with Down syndrome and how this compares to typical development.

## Predictors of lexical development in Down syndrome

Following the model of holistic development (Fröhlich & Haupt, 2004; Van Balkom, 2009, see Figure 1), the lexical development of children is interrelated with several other developmental domains, such as cognition, memory, attention, adaptive level of functioning and socio-emotional skills. Given that word learning in children with Down syndrome seems to be delayed rather than deviant from typical development (Polisenka, & Kapalková, 2014), the same predictors that play a role in typical development (see Chapter 4) may also play this role in the development of receptive and expressive vocabulary in children with Down syndrome. Most of the vocabulary studies in children with Down syndrome used a cross-sectional design and only focussed on a limited number of predictors. Only a few studies have used longitudinal designs to assess the vocabulary development of children with Down syndrome, due to which there is a lack of data on trends in vocabulary development (Zampini & D'Odorico, 2013). Insofar longitudinal studies on vocabulary development have been conducted only a limited number of predictors was incorporated (e.g., Berglund et al., 2001; Miller, 1999; Oliver & Buckley, 1994).

Recently, Næss, Lervag, Lyster and Hulme (2015) followed the receptive and expressive vocabulary development of a cohort of 43 6-year-old children with DS. Although the longitudinal design and the inclusion of a control group of 57 3-year-old typically developing children matched on nonverbal mental abilities are strengths of Naess et al.'s study, it also has a major weakness. Measures

included in the study to predict vocabulary development were only verbal short-term memory tasks (i.e., word span, sentence memory, and non-word repetition) and early vocabulary levels as measured by the British Picture Vocabulary Scale (BPVS-II; Dunn, Dunn, Whetton, & Burley, 1997) and Picture Naming (WPPSI-III; Wechsler, 2002), not looking at many of other possible predictors such as the developmental domains discussed earlier. Næss et al. (2015) concluded that later vocabulary skills could not be significantly predicted by both early vocabulary measures and verbal short-term memory. To date, researchers in the field of DS thus show a one-sided focus on only a few predictors of the lexical development in DS. Also, none of these studies have taken the communicative perspective into account, leaving us with only little insight in the lexical development of children with Down syndrome and its predictors.

## Identifying individual pathways in lexical development

Although identifying precursors of lexical development on the group level is of high importance, "genetic disorders do not have uniform effects on every individual, can change their effects due to developmental and environmental effects, and must not be considered the sum total of any person's overall genetic endowment" (Hodapp, 1997, p.68). This indicates that not every child with Down syndrome necessarily shows all etiology-specific behaviours, and that the behavioural phenotype is not per se useful in clinical-decision making in interventions regarding lexical development for children with Down syndrome. Down syndrome has been extensively described at the group level, due to a focus on behavioural phenotyping, downplaying individual variation and treating Down syndrome as a homogeneous group. In order to fully understand how children with Down syndrome develop their vocabulary, it is crucial to study how individual differences in underlying processes, such as attention and motor control, constrain higher-level cognitive outcomes, such as language and vocabulary development (Karmiloff-Smith et al., 2016). An important challenge is to identify the causes of individual differences in word learning in Down syndrome, "given their (i.e., individual differences) importance in developing a true theory of language development as well as their importance for assessment and intervention" (Galeote, Sebastián, Checa, Rey, & Soto, 2011). Individuals with Down syndrome would benefit more from interventions that are tailored to their own critical areas of strengths and weaknesses (Neil & Jones, 2016).

To realize the full benefit of interventions, such as AAC, for children with Down syndrome, a comprehensive assessment is required to make appropriate recommendations and decisions regarding the particular communication and language interventions to be implemented (Beukelman & Mirenda, 2013).

However, little is known about how clinicians make these decisions. It is thought that most practitioners base their decisions on clinical reasoning from experience (Schlosser & Raghavendra, 2004). This may be due to limited practical guidelines for language and communication assessment and decision making (Dietz, Quach, Lund, & McKelvey, 2012). In agreement with Rowland, Fried-Oken, Steiner, Lollar, Phelps, Simeonsson and Granlund (2012), there are no theoretically and clinically driven models to help integrate information about the myriad of underlying and contributing factors that may affect communicative and language competence and performance, including severe health conditions, functional impairments, environmental and social network barriers. The International Classification of Functioning, Disability, and Health (ICF) may provide a framework to determine these underlying and contributing factors.

The World Health Organization (WHO) has developed the ICF and a version for Children and Youth (ICF-CY). These are sophisticated classification systems that provide an interdisciplinary international language of health and health-related issues for all people across the lifespan (WHO, 2001, 2007). By introducing the ICF framework, a transition is seen from an emphasis on disability to an emphasis on health and wellness (Üstün, Chatterji, Bickenbach, Kastanjsek, & Schnieder, 2003). There are two parts to the ICF and ICF-CY, each containing two components. Part 1: Functioning and Disability contains the components of (a) Body Functions and Structures and (b) Activities and Participation. Part 2: Contextual Factors, contains the components of (a) Environmental Factors and (b) Personal Factors. An overview of all components and domains within the ICF and ICF-CY can be found in Table 1. Body functions are "the physiological functions of body systems (including psychological functions)". Body structures are "anatomical parts of the body such as organs, limbs and their components" (p. 10). Activity is "the execution of a task or action by an individual", while Participation is the "involvement in a life situation" (WHO, 2001, p. 10). Environmental factors include the persons' environment, availability of resources and support. Personal factors include the attributes of the person, and the internal influences on functioning and disability, such as gender, age, coping style, social background, education, past experiences, and temperament and character style.

The ICF and ICF-CY are classification systems and are not assessment or intervention tools (Threats & Worrall, 2004), but the insight gained from using the framework enables consideration of holistic assessment and intervention practices. Consideration of all components (see Figure 1) should be interwoven within an intervention approach (McLeod, 2006). This way individual predictors of lexical development can be studied in children with Down syndrome and interventions can be planned accordingly.

Table 1. *Components and Domains within the ICF-CY.*

| **Body functions** | **Body structures** |
|---|---|
| Mental functions | Structures of the nervous system |
| Sensory functions and pain | The eye, ear, and related structures |
| Voice and speech functions | Structures involved in voice and speech |
| Functions of the cardiovascular, haematological, immunological and respiratory systems | Structures of the cardiovascular, immunological and respiratory systems |
| Functions of the digestive, metabolic and endocrine systems | Structures related to the digestive, metabolic, and endocrine systems |
| Genitourinary and reproductive functions | Structures related to the genitourinary and reproductive systems |
| Neuromusculoskeletal and movement-related functions | Structures related to movement |
| Functions of the skin and related structures | Skin and related structures |
| **Activities and Participation** | **Environmental factors** |
| Learning and applying knowledge | Products and technology |
| General tasks and demands | Natural environment and human-made changes to environment |
| Communication | Support and relationships |
| Mobility | Attitudes |
| Self-care | Services, systems and policies |
| Domestic life | |
| Interpersonal interactions and relationships | |
| Major life areas | |
| Community, social, and civic life | |

# The present research project

## Research questions

The aim of the present research project was to provide insight in the lexical development of receptive and expressive vocabulary of young children with Down syndrome. In short, studies addressed three research questions:

1. What is the validity of the Dutch version of the MacArthur-Bates Communicative Development Inventories measuring lexical development both in spoken and signed modalities in children with Down syndrome?
2. Which words do children with Down syndrome use most in their spontaneous language production?
3. What are the predictors of lexical development in young children with Down syndrome at a group level and how can strengths and weaknesses in these language and communication-related developmental domains be identified in individual cases?

In order to answer the first research question a longitudinal study was performed to investigate both the concurrent and predictive validity of a parental report on vocabulary size.The second question was answered by conducting a cross-sectional study wherein spontaneous language samples were recorded in several settings and with multiple communication partners in order to obtain language samples mirroring a wide range of language use. The third question was examined in two separate studies. The predictors of lexical development at a group level were identified within a longitudinal design. Individual strengths and weaknesses in child- and environmental related predictors were studied within a multiple-case study design.

## Outline of the dissertation

The next four chapters each represent an empirical research paper accepted or submitted for publication. In Chapter 2 (‘The validity of measuring lexical development in children with Down syndrome by parental reports’), the concurrent and predictive validity of an adapted version of the N-CDI is examined. The N-CDI was adapted to both include spoken and signed modalities for the assessment of lexical size.

In Chapter 3 (‘Functional word use of children with Down syndrome’), spontaneous language samples of young children with Down syndrome were explored in order to investigate their functional word use in multiple communicative settings and with several communication partners.

Chapter 4 ('Predictors of lexical development in children with Down syndrome') represents a longitudinal study in which the lexical development over a 1.5-year period of children with Down syndrome was examined. The predictive role of child- and environment-related factors in the lexical development of these children is explored.

In Chapter 5 ('A comprehensive assessment of lexical development in Down syndrome: Integrative profile of communication performance') a multiple-case study design is used to show individual pathways of lexical development in children with Down syndrome. Individual, child- and environment-related strengths and weaknesses are determined within the ICF-CY framework to direct clinical-decision making in language and communication interventions.

Chapter 6 provides a summary of the results of the four studies, followed by theoretical implications. Ultimately, directions for future research, and practical implications are discussed.

# Chapter 2

## The validity of measuring lexical development in children with Down syndrome by parental reports

This chapter is based on Deckers, S. R. J. M., Van Zaalen, Y., Mens, E. J. M., Van Balkom, H., & Verhoeven, L. (2016). The concurrent and predictive validity of the Dutch version of the Communicative Development Inventories in children with Down Syndrome for the assessment of expressive vocabulary in verbal and signed modalities. Research in Developmental Disabilities, 56, 99-107.

## Abstract

The expressive vocabulary of children with Down syndrome is generally measured with parental reports, such as the McArthur-Bates Communicative Development Inventories (CDI), given that standardized tests for assessing vocabulary levels may be too difficult for most young children with Down syndrome. The CDI provides important insight into the parents' perception of their child's vocabulary development. The CDI has proven to be a valid measurement of expressive vocabulary, spoken and gestural, in typical and atypical populations. The validity in children with Down syndrome is not well established and signed vocabulary is often not included. This longitudinal study examined the concurrent and predictive validity of the Dutch version of the CDI (N-CDI) in children with Down syndrome between 2;0 and 7;6 years old to assess spoken and signed vocabulary. N-CDI scores were assessed on strength of association with mental age, an expressive vocabulary test and spontaneous language analyses in a play setting with parents at T1 and T2 (1.5 years later), and a therapy setting with speech language pathologists at T1. The results of the present study show that the N-CDI is a valuable and valid measurement of expressive vocabulary in children with Down syndrome. Strengths and weaknesses of several assessment methods for expressive vocabulary are discussed.

## The validity of measuring lexical development in children with Down syndrome by parental reports



Many children with Down syndrome show a significantly delayed onset of the production of their first words, despite them having normal babbling patterns (Naess, Lyster, Hulme & Melby-Lervag, 2011; Stoel-Gammon, 1997). Smith and Oller (1981) found that children with Down syndrome have a delayed onset of meaningful first words and that the majority of utterances made by them were not meaningful, even after the appearance of their first intelligible words (Smith & Oller, 1981; Stoel-Gammon, 1997). Although Stoel-Gammon (2001) claims that children with Down syndrome produce their first spoken word around the average age of 1;9 years, Gillham (1990) determined that word onset in children with Down syndrome is possibly not until the age of 3;9 years. Berglund et al. (2001) used a Swedish version of the MacArthur-Bates Communicative Development Inventories (CDI) in a large sample of 330 children with Down syndrome between one and five years old. They found that some of them started to talk around their first birthday while others had not even started at the age of five, showing large individual differences in vocabulary development.

Contextual factors, attention span of the child, and lengthiness of data acquisition are less likely to influence assessment via parental reports, of which the CDI is the most often used both in research as in clinical practice. For the speech language pathologist, these parental reports are cost-effective and administration is minimally intrusive. Parents may have a good understanding of their child's vocabulary use, because they observe and interact with their child in several contexts on a daily basis (Feldman et al., 2005; Pan et al., 2004), and in the case of children with Down syndrome, their parents may understand imprecise articulated speech or signs of their child better. The particular moment of the word produced in time is not relevant, and parents may take days to fill out the forms. This means that words that are produced days before, and remembered by the parents, are included in the list as well. The CDI provides important insight into the parents' perception of their child's vocabulary development. The data obtained via parental report is not limited by certain factors that could have affected the child's performance on tests, such as fatigue or motivation (Miller et al., 1995). The usefulness of these parental reports is based on the representative and extensive knowledge of parents regarding their children's language skills (Dale & Goodman, 2005).

Several studies showed the CDI to be highly correlated with more structured vocabulary measures in typically developing children such as the Expressive One Word Picture Vocabulary Test (EOWPVT), the Bayley Scales of Infant

Development and the Peabody Picture Vocabulary Test (Jackson-Maldonado, Thal, Marchman, Bates, & Gutierrez-Clellen, 1993; Fenson et al., 2007; Pérez-Pereira & Resches, 2011). In typically developing children, multiple studies have proven the CDI to be concurrently and predictively valid in several languages (e.g., Deniz Can et al., 2012; Feldman et al., 2005; Lee, 2011a; Pérez-Pereira & Resches, 2011). But, there are some studies that have questioned the accuracy of the CDI, particularly in atypical groups, in which parents tend to over- or underestimate the vocabulary use of their child (Roberts et al., 1998). However, the CDI proved to be valid for several of these groups, such as toddlers in low-income families (Pan, Rowe, Spier & Tamis-Lemonda, 2004), children at risk for dyslexia (Koster et al., 2005), late talkers (Klee, Pearce, & Carson, 2000), and children who stutter (Ratner & Silverman, 2000). The validity of the CDI for children with Down syndrome has not been investigated that extensively. The accurate reporting of language abilities for parents of these children could be even more challenging due to the asynchronies of children with Down syndrome within different linguistic domains and sub-domains, as well as in other developmental areas, as often found in Down syndrome (Abbeduto, Warren & Conners, 2007). Also, many children with Down syndrome have difficulty producing verbal words consistently (Kumin, 2003), which may compromise the number of intelligible words understood by parents.

The study of Miller, Sedey and Miolo (1995) was the first to examine the concurrent and predictive validity of the CDI in children with Down syndrome. To examine the concurrent validity, results from the CDI of 44 children with Down syndrome, and 46 typically developing children, with mental ages from 12 to 27 months, were compared to laboratory measures of vocabulary size. Laboratory measures contained a language subset of the Bayley Scales of Infant Development (BSID) and observations of 15 minutes of free play with a researcher who was unknown to the child and 15 minutes of free play with the parent. Miller et al. (1995) found high correlations between CDI scores and both the observed vocabulary ($r = .82$) and BSID scores ($r = .77$).The predictive validity was examined for 20 children with DS by obtaining correlations between these measurements around 20 months mental age (Time A) and around 28 months mental age (Time B). They found moderate correlations around .60 between the CDI scores at Time A and CDI scores, observed vocabulary and BSID scores at Time B. Although the Miller et al. (1995) study provides important insights into the concurrent and predictive validity of the CDI in children with Down syndrome, the study has it weaknesses. Only observations of the children's language in an unknown laboratory setting with an unknown researcher and with a parent were used, possibly evoking less utterances than the child would have in

a familiar setting (Mervis & Becerra, 2003). Also, a preliminary version of the CDI was used and only spoken words were counted and not signed vocabulary. Investigating only spoken vocabulary, and not manual vocabulary (signs and/or gestures), in an unfamiliar setting, may have led to the underestimation of the total vocabulary size, both in the CDI as well as in the spontaneous language sampling. In an Italian study, Zampini and D'Odorico (2012) studied the validity of the Italian version of the CDI (i.e., Questionnario Il Primo Vocabulario del Bambino) to determine the expressive vocabulary size of 48 children with Down syndrome between 2 and 5 years of age, and found an association with developmental age and gender, with girls showing an advantage. Only spoken vocabulary was included in the analyses.

Most studies of vocabulary development in children with Down syndrome show this important limitation. The production of words is often taken into account only in its vocal modality, whereas many children with Down syndrome spontaneously produce numerous gestures and signs (Galeote et al., 2008), which should also be considered in the assessment of vocabulary development (Berglund et al., 2001). Bello et al. (2014) investigated the production of nouns and predicates in fourteen children with Down syndrome with a developmental age of 34 months. Next to spoken vocabularies, they also included gestural production (i.e., the use of co-speech gestures), and coded deictic gestures (e.g. pointing) and representational or symbolic gestures. Representational and symbolic gestures can be defined as those gestures that substitute specific lexical items, for example closing the hand with tight fingers and moving it towards the mouth to simulate "to eat".

Gesture production is considered a strength of children with Down syndrome compared to their vocal language skills and also compared to typically developing peers (Galeote et al., 2011). Galeote et al. (2008) studied the relationship between cognitive development and vocabulary size in both the vocal and gestural modality in 66 Spanish children with Down syndrome. They adapted the CDI and added a column for *understands and gestures* in which the gestures represented specific lexical items (i.e. representional gestures). Children with Down syndrome were found to have comparable vocabulary sizes to those of typically developing peers. Galeote et al. (2011) also used the adapted CDI for gestures, and compared the spoken and gestured vocabulary of 186 children with Down syndrome with a mental age between 9 and 29 months with that of typically developing peers. Zampini and D'Odorico (2009) also showed the validity of the production section (i.e. spoken words and gestures) of the Italian version of the CDI for children with Down syndrome and found that gesture production at 36 months was significantly correlated to the subsequent vocabulary production, assessed at 42 months. From these studies it can be

concluded that the CDI seems to be a valid measurement of vocabulary size in children with Down syndrome.

All these studies investigated the use of gestures, next to speech, in children with Down syndrome. However, it is recognized that interventions using Augmentative and Alternative Communication (AAC) strategies may help children with Down syndrome communicate earlier and more successfully. A large number of young children with Down syndrome are ready to communicate with a language system such as sign language by ten to twelve months of age, which requires less advanced neuromuscular skills than speech (Kumin, 2003). A poor intelligibility of speech and the significantly delayed onset of spoken words are two primary reasons to introduce the use of signs to children with Down syndrome (Brady, 2008). As a sign is conveyed in the visual-motor modality and because infants use communicative gestures prelinguistically, children learning signed languages may have a distinct advantage over children solely learning spoken language, especially in the acquisition of first words (Anderson & Reilly, 2002). The combined use of signs or gestures with speech facilitates communication development. Wright, Kaiser, Reikowsky and Roberts (2013) showed that a naturalistic sign intervention including signs and spoken words appeared to be more effective on expressive language skills in children with Down syndrome than interventions that taught spoken words alone.

Many other studies also mention the use of signs as a strength in children with Down syndrome when compared to individuals with a different etiology of their intellectual disability (Attwood et al., 1988; Caselli et al., 1998; Naess et al., 2011; Roberts et al., 2007). As high as 87% of the parents of 330 children with DS between the ages of one and 5;6 years reported their children using speech and signs simultaneously (Berglund et al., 2001). The spoken vocabulary sizes, as reported by Berglund et al. (2001), may thus be an underestimation of the actual vocabulary size of children with Down syndrome.

Examples of AAC intervention strategies in clinical practice are the Signed Exact English System (SEE) for English and Nederlands Ondersteund met Gebaren {Sign Supported Dutch} (NmG), which are spoken languages in a visual mode, supporting the spoken word in the visual modality. In Dutch clinical practice, the use of NmG in young children with Down syndrome is a common intervention strategy, as a result of which these children use signs as an important mode of communication. By the age of five years, children with Down syndrome have usually outgrown the need to use signs as their primary communication system due to an increased intelligibility of their speech, unless they have a significant hearing loss or verbal apraxia or dysarthria, and the use will gradually fade out over the next year(s) (Kumin, 2003). The signs used in

NmG stem from Dutch Sign Language, and are therefore actual signs and not just deictic or representational gestures. Assessment of vocabulary size, using the CDI, in these children with Down syndrome should therefore also incorporate their manual sign vocabulary size.

Zink and Lejaegere (2002) tested the concurrent and predictive validity of the Dutch version of the CDI (N-CDI, in which the N stands for Netherlands) in typically developing Dutch children and found the validity to be moderate to high. The goal of the present study was to adapt the N-CDI to also recognize the role of signs in the vocabulary size of Dutch children with Down syndrome. The concurrent and predictive validity of this adapted version of the N-CDI in Dutch children with Down syndrome were investigated by assessing the vocabulary size via parental reports, by standardized tests and in spontaneous language samples. Based on earlier validity studies of other forms of the CDI, high correlations between all these measurements are hypothesised.

## Method

### Participants

Parents and their children with Down syndrome were recruited for this longitudinal study from special Down syndrome teams in hospitals in the Netherlands, through *Stichting Downsyndroom*, the Dutch association for parents with a child with Down syndrome, and through speech language pathologists in clinical practice. Because the Berglund et al. (2001) study showed some Down syndrome children had not started speech development at age five, it was decided to include children with a broader age range. Parents were included in the study if their child met the following inclusion criteria: (a) chronological age between 2;0 and 7;6 years, (b) Dutch-speaking home environment, and (c) children were all genetically diagnosed with DS (trisomy 21). The present study was approved by an Institutional Review Board (NL38926.091.12).

Twenty-five children with Down syndrome participated in the present study: 13 boys and 12 girls. At the start of the study, the participants had a mean chronological age of 55 months (*SD = 17; range*: 29-87) and a mean mental age of adaptive functioning of 23 months (*SD* = 8; *range*: 10-40). All children came from Dutch-speaking households, however, the parents of two children had a Turkish background. Sixteen of the children visited schools or daycare centers for regular education, ten children visited schools or daycare centers for special education.

## Materials and procedures

The N-CDI: Words and Sentences (N-CDI; Zink & Lejaegere, 2002) was used to determine the size of expressive vocabulary as indicated by the parents. The Words and Sentences form of the N-CDI (for children with a chronological or mental age of 16 – 30 months) contains a checklist of 702 words divided into several categories, including both content and function words. The Dutch Words and Sentences form was chosen over the Dutch Words and Gestures forms given that: (a) the words assessed in the Dutch Words and Gestures form are all included in the Dutch Words and Sentences form, where the vocabulary described in the Words and Sentences form is larger; (b) in order to compare the vocabularies of children in the present study, the same measurement of expressive vocabulary should be used at both time points; and (c) given the longitudinal design of the present study it was to be expected that the mental ages of children would increase, due to which ceiling levels on the Words and Gestures form would have been reached by many of the children.

The parents of the children were asked to fill out this parental form prior to video recordings of spontaneous language samples of their children. In the present study, the N-CDI form was adapted so that parents could indicate vocally produced words as well as signed words. For this, the adapted version of the CDI for American Sign Language (ASL-CDI) to assess sign language skills in children ages 8 to 36 months was explored, which has demonstrated excellent reliability and validity (Anderson & Reilly, 2002). For the present study, a new column was added to the N-CDI. A first column could be used to indicate which words were verbally produced by the child, a second column could be used to indicate which words were produced as a sign. Parents could also mark both columns, indicating that a particular words is produced by their child both in the spoken and signed modality.

The total number of unique words used by the children was used for further analyses. This total number was the sum of the words that were spoken and those that were produced only through signs. A word that children uttered both verbally and signed was counted as one. The N-CDI form only allows a verb to be checked once, irrespective of which form of the verb is produced by the child. Thus, when a child produces give, gives, and gave, this counts only as one unique word for the verb give. The same way of counting vocabulary size was used in the spontaneous language sampling.

Parents were asked to fill out the N-CDI at home twice (at T1 and T2) and to return the lists by mail. Maldonado et al. (1993) found no significant differences in how parents filled out the CDI, when they compared a setting in which parents received and returned the CDI via mail or when personal interviews were

conducted, indicating that filling out the N-CDI at their home environment yields equally reliable information about vocabulary size. Between T1 and T2 was a period of 1;6 years.

Mental age of the participants were obtained both at T1 and T2. In clinical practice and research settings, the Bayley Scales of Infant Development-III is currently the most commonly applied measurement in the assessment of early development both, but the scale seriously underestimates developmental delay in 2-year-old children (Anderson, De Luca, Hutchinson, Roberts, & Doyle, 2010). Provided the described problems with standardized tests in the introduction, it was chosen to measure the level of adaptive functioning with a parental questionnaire as well. The level of adaptive functioning can be defined as the collection of conceptual, social and practical skills that have been learned by individuals in order to function in their everyday live (American Association on Intellectual and Developmental Disabilities, 2016), and can be measured by the *Vineland Screener 0-6 years* (VS; Van Duijn et al*.,* 2009). The VS consists of 90 items adhering to four components of adaptive functioning: communication, daily living skills, socialization and motor skills. The reliability and validity of the Dutch version of the Vineland Screener were found to be high in typically developing children (Van Duijn, Dijkxhoorn, Noens, Scholte, & Van Berckelaer-Onnes, 2009). Van Duijn, Dijkxhoorn, Scholte and Van Berckelaer-Onnes (2010) used the Vineland screener to determine the level of adaptive functioning of 984 Dutch children with Down syndrome, aged between 0 and 12 years, in comparison to the level in typically developing peers. They concluded that children with Down syndrome acquired skills in a similar sequence and according to a similar trajectory. The Vineland Screener is therefore a valid measurement of the level of adaptive functioning in children with Down syndrome and can be used to determine a mental age of adaptive functioning.

To determine the size of expressive vocabulary in spontaneous language samples, video recordings were examined. These video recordings existed of play settings between the child and a parent in the home environment, and the child and a speech language pathologist (SLP) in the therapy environment. A spontaneous language analysis was done on the basis of 45 minutes of spontaneous language use per child: 15 minutes with a SLP (therapy setting), and 15 minutes in a play setting with parents at T1 and 15 minutes in a play setting with parents at T2. The session with speech therapists was also planned for T2, however, only two speech therapists were willing to participate again at that moment. Due to a lack of data, this setting had to be excluded from the analyses. Videotape-recordings were verbatim transcribed and the number of intelligible utterances made by the children, either spoken or signed, were analysed and counted in these various

settings. All unique words, either spoken and/or signed, were counted. Words were only counted if they were produced spontaneously and functionally by the child and not when it was only an imitation or repetition of a word produced by parents or SLPs. The total number of unique words used by the children was used for further analyses. This total number was the sum of the words that were spoken and those that were produced only through signs. A word that children uttered both verbally and signed was counted as one.

To ensure reliability of the spontaneous language analysis, videos were also transcribed by a second, independent researcher who had not seen the previously made transcriptions, who was unaware of the research questions and of the children's N-CDI scores. The principles of inter-judge agreement were applied, which can be used to obtain reliable transcripts of utterances in individuals who have limited or a high level of unintelligible speech (Kovacs & Hill, 2015). Complete inter-judge agreement is obtained by independently transcribing and analyzing a language sample and then resolving discrepancies by consensus as needed (Hegde, 2003). It is typically 100%, unless the raters are unable to reach a consensus (Kovacs & Hill, 2015). Small differences that were found in the transcripts were reconsidered and after consensus altered in the transcriptions.

After the recordings of the play settings with parents, the EOWPVT (Brownell, 2000) was administered, both at T1 and T2, to assess expressive vocabulary size in a standardized way. Both verbal and signed responses were taken into account.

## Statistical Analysis

To test the concurrent validity, Spearman's Rho correlation were determined ($\alpha < .05$) between the N-CDI vocabulary size, the mental age and the three other vocabulary measures: (a) EOWPVT scores, (b) the number of different (i.e. unique) words produced spontaneously during a 15-minute play setting with a parent, and (c) the number of different (i.e. unique) words produced spontaneously during a 15-minute therapy setting with a known SLP. This was done for both T1 and T2, except for the therapy setting at T2. To test the predictive validity of the N-CDI in children with DS, the N-CDI scores at T1 were correlated (Spearman's Rho; $\alpha < .05$) with the vocabulary measures obtained at T2: (a) the N-CDI vocabulary size, (b) the EOWPVT, and (c) the number of different (i.e. unique) words produced spontaneously during a 15-minute play setting with a parent. Correlations are interpreted accordingly the rules of thumb as presented in Mukaka (2012): .90 – 1.00 is very high, .70 - .90 is high, .50 - .70 is moderate, .30 - .50 is low/weak.

## Results

A wide range of parent reported vocabulary sizes was evident. The total vocabulary size according to the N-CDI ranged from 7 to 697 words at T1 ($N = 24$), and from 7 to 623 words at T2 ($N = 18$). One child, with the highest mental age of 40 months, reached N-CDI ceiling levels at T1 due to which parents did not receive a request to fill out the N-CDI at T2. The mean scores, standard deviations, and ranges for N-CDI scores, EOWPVT scores and observed vocabulary are reported in Table 1. One parent did not return the N-CDI form at T1, and EOWPVT administration could not be scheduled provided medical constraints in two cases. Only children that had N-CDI scores at T1 were included at T2. Most parents indicated on the N-CDI forms that their children uttered more words at T2 than at T1 (see Table 1). However, three parents marked a smaller amount of words at T2 (range 1-49 words). The number of spoken and signed words are also included in Table 1. The mean number of spoken words almost doubled over time, while the mean number of signed words showed a small decrease, although the range indicates that some children used more signed words at T2 than at T1. From Table 1, it is also obvious that at least one child had not begun to speak at T1 and still did not speak at T2. When looking at the EOWPVT scores, about half of the children uttered more words at T2 compared to T1, six children scored the same amount at both time points (i.e, five of them scored a zero), and four children uttered a lower amount of words at T2 compared to T1 (range 1-10 words). The mean number of words uttered in therapy setting and play setting with parents at T1 does not differ significantly. The mean number of words uttered during play situations with parents at T2 is almost twice the size of those at T1. Most children uttered more words during play with parents at T2 than during T1, as only five children used fewer words (range 4-14 words less).

Table 1. *Results of N-CDI, EOWVPT and Observed Vocabulary.*

| | T1 | | | T2 | | |
|---|---|---|---|---|---|---|
| **Variable** | **N** | **Range** | **Mean ± SD** | **N** | **Range** | **Mean ± SD** |
| N-CDI Total vocabulary | 24 | 7 - 697 | 215.3 ± 178,8 | 18 | 7 - 623 | 307.0 ± 180.0 |
| N-CDI Verbal vocabulary | 24 | 0 - 697 | 155.5 ± 203.3 | 18 | 0 - 623 | 304.9 ± 196.0 |
| N-CDI Signed vocabulary | 24 | 0 - 301 | 91.9 ± 85.9 | 18 | 0 - 351 | 84.1 ± 118 |
| EOWPVT | 23 | 0 - 38 | 12.2 ± 11.1 | 24 | 0 - 48 | 18.2 ± 13.6 |
| Words observed with parents | 25 | 0 - 48 | 19.3 ± 13.7 | 24 | 3 - 104 | 38.5 ± 27.2 |
| Words observed with SLP | 20 | 0 - 42 | 19.4 ± 14.3 | | | |

## Concurrent validity

Significant positive correlations between N-CDI scores and the validation measures were obtained for both T1 and T2 (see Table 2). All concurrent validity measures both at T1 and T2 were significantly correlated with N-CDI scores. At T1, the parent report vocabulary, as measured by the N-CDI, showed a *high correlation* with (a) mental age, (b) EOWPVT scores, and (c) the number of different words the children with DS produced during therapy settings with SLPs. The correlation between the N-CDI and the number of different words produced during play settings with parents was found to be moderate. At T2, correlations between N-CDI scores and the three measurements for concurrent validity were also found to be moderate. Based on these correlations the concurrent validity of the adapted N-CDI in Dutch children with DS is moderate to high.

## Predictive validity

The statistically significant positive correlations between N-CDI scores at T1, N-CDI scores, EOWPVT scores and observed vocabulary size in a play setting with parents at T2 are reported in Table 2. A high correlation between N-CDI scores at T1 and T2 was found. Moderate correlations were detected between N-CDI scores at T1 and EOWPVT scores or words observed with parents at T2. Based on these correlations the predictive validity of the adapted N-CDI in children with DS is moderate to high.

Table 2. *Spearman Correlations of N-CDI at T1 and T2 with validity measurements.*

| | N-CDI T1 | N | N-CDI T2 | N |
|---|---|---|---|---|
| **Concurrent validity** | | | | |
| Mental Age | .87** | 24 | .66** | 18 |
| EOWPVT | .85** | 22 | .61** | 17 |
| Words Observed with Therapist | .81** | 19 | | |
| Words Observed with Parents | .58** | 23 | .70** | 16 |
| **Predictive validity** | | | | |
| N-CDI | .73** | 18 | | |
| EOWPVT | .54** | 22 | | |
| Words Observed with Parents | .45* | 22 | | |

*. Correlation is significant at the 0.05 level (2-tailed).

**. Correlation is significant at the 0.01 level (2-tailed).

## Discussion

The goal of the present study was to examine the concurrent and predictive validity of the adapted version of the N-CDI in Dutch children with Down syndrome in order to establish the utility of the N-CDI in determining expressive vocabulary size (both spoken and signed words). Since the N-CDI scores in the present study were found to show moderate to high correlations with all validity measures, the adapted N-CDI seems to be a valuable and valid tool to assess the expressive vocabulary size in children with Down syndrome, both taking into account verbal and signed vocabulary.

There are some studies that have questioned the accuracy of the CDI, particularly in atypical groups such as Down syndrome, in which parents tend to over- or underestimate the vocabulary use of their child (Roberts et al., 1998). The results of the present study overall indicate that parents can accurately fill in the N-CDI, given the high correlations with both the EOWPVT and the number of words spontaneously produced in a therapy setting with SLPs and a play setting with parents. Although spontaneous language analysis gives a precise overview of the expressive vocabulary used in a particular context, other words could have been found when other contexts were analysed. For example, an observation of the child playing in the sandbox will not provoke the same words that are related to baking cookies. On the other hand, it is known that children's language production is influenced by the communication style of their communication partner. Partners that adjust their language to the language production of the child evoke and prompt more and more variant language in comparison to those communication partners who do not adjust their production to the language production level of the child. Different communicative partners will therefore evoke different words and sentences (Pepper & Weitzman, 2004). Thus, assessing the vocabulary size on the basis of spontaneous language analyses is a lengthy and time-consuming effort, which is often not possible for SLPs in clinical practice. The N-CDI provides important insights in the whole range of words that children with DS use in different contexts and with different communication partners.

Contextual factors, attention span of the child, and lengthiness of data acquisition are less likely to influence assessment via parental report. For the SLP, these parental reports are cost-effective and administration is minimally intrusive. Parents may have a good understanding of their child's vocabulary use, because they observe and interact with their child in several contexts on a daily basis (Feldman et al., 2005; Pan et al., 2004), and in the case of children with DS, they may be better able in understanding imprecisely articulated speech and signs of their child. The particular moment of the word produced in

time is not relevant, in comparison to measuring vocabulary in a standardized assessment.

A couple of remarkable differences were observed in the assessment results, which will be discussed in further detail. Firstly, only half of the children uttered more unique words on the EOWPVT at T2 than at T1. Some of the other children scored equal to T1 and some children uttered fewer correct words at T2. This is most likely due to variables that could have affected the child's performance, such as fatigue or motivation (Miller et al., 1995). If all children cooperate in the test setting and therefore show their actual skills, a high correlation is expected to be found between N-CDI scores and EOWPVT scores. However, in young children, test results are likely to be an underrepresentation of the child's abilities (Feldman et al., 2005), which may certainly be true in children with Down syndrome.

Secondly, although the results of the present study show the N-CDI to be a valid measure of expressive vocabulary in children with Down syndrome, there are some findings that compromise the reliability. Three out of 25 parents indicated that their children uttered fewer words at T2 in comparison to T1 with a range up to 49 words. Also, a few parents indicated fewer words on the N-CDI than their child uttered during the assessment. This may be due to the earlier mentioned tendency for parents of children from atypical populations, such as Down syndrome, to over- or underestimate the vocabulary use of their child (Roberts et al., 1998). However, given the high correlations with multiple validity measures, this tendency does not seem to compromise the results of the present study. Another possibility is a more clinical observation. There were some children at T1 that relied almost exclusively on the use of signs, but at T2 relied most on speech. The number of words used do not necessarily increase largely or may even decrease for some children, but the mode in which they are expressed does. Some children with Down syndrome seem to stop using signs at the moment that they are able to express themselves well enough through speech. This, however, might mean that they express fewer words overall. A large number of young children with Down syndrome are ready to communicate with signs by ten to twelve months of age, which require less advanced neuromuscular skills than speech (Kumin, 2003). There seems to be a close link between motor programs associated to actions, gestures and signs, and spoken linguistic representations in children (Capirci, Caselli, & De Angelis, 2010). Our observation that children switch their primary mode of communication from signs to speech is in accordance with observations of Kumin (2003) who found that children with Down syndrome, around the age of five years, have usually outgrown the need to use signs as their primary communication system and the use will gradually fade out in the next year(s).

Since it was not the focus of the present study, this particular use of either spoken or signed words was not analysed.

Future, longitudinal studies should target the use of speech or signs for specific words, using the adapted version of the N-CDI from the present study, in order to gain more insight in the qualitative development of vocabulary in children with Down syndrome. Given the wide use of signs by children with Down syndrome in the present study, following Galeote et al. (2008), it would be beneficial to explain to parents that sign instruction in the early stages of language development helps to improve initial communication of children with Down syndrome.



## Limitations

The present study has some limitations. The sample size is relatively low, with only 25 children and parents participating in the present study. Although the recruitment of young children with Down syndrome and their parents is not easy, and small sample sizes are common in research with children with Down syndrome, this restriction may limit the generalisability of the findings of this study. The children in the present study showed a range in chronological age of 58 months, a range in mental age of 30 months, and a wide range in the expressive vocabulary size, both in words and in signs, indicating large individual differences. These differences are found to be common among children with Down syndrome in their vocabulary development (Galeote et al., 2008). Also, it would have benefitted the usability of the adapted N-CDI to investigate the reliability. Given the small sample size, all parents should have filled out the N-CDI twice within a small period of time. Most parents indicated that this would take them too much time, due to which it was decided not to test the reliability. However, there is abundant evidence for the reliability of other forms of the CDI, also in adapted versions which included signs (Anderson & Reilly, 2002). It it hypothesised that reliability should not be an issue, but this should be investigated in a future study. Finally, both the N-CDI and the Vineland Screener for adaptive level of functioning were filled out by the parents of the children. It could have been that the children's perceived abilities are over- or underestimated by both of the measures. Provided the high correlations of the N-CDI with spontaneous language samples found in the present study and the Vineland Screener showed to be valid for measuring the mental age in children with Down syndrome (Van Duyn et al., 2010), parents of children with Down syndrome in the present seem to perceive the abilities of their children adequately.

## Conclusion

To conclude, the present study showed that the adapted N-CDI is a fast, efficient and valid instrument to determine the expressive vocabulary size in children with Down syndrome. Furthermore, as the use of signs is highly frequent in children with Down syndrome, the number of uniquely signed words should be included in the assessment of vocabulary size. The N-CDI, as well as expressive vocabulary tests as the EOWPVT, can be easily adapted to also include signs or other Augmentative and Alternative Communication.

# Chapter 3

## Functional word use of children with Down syndrome

This chapter is based on Deckers, S. R. J. M., Van Zaalen, Y., Van Balkom, H., & Verhoeven, L. (2017). Core vocabulary of young children with Down syndrome. Augmentative and Alternative Communication, 33, 77-86.

## Abstract

The aim of this study was to develop a core vocabulary list for young children with intellectual disabilities between 2 and 7 years of age because data from this population are lacking in core vocabulary literature. Children with Down syndrome are considered one of the most valid reference groups for researching developmental patterns in children with intellectual disabilities; therefore, spontaneous language samples of 30 Dutch children with Down syndrome were collected during three different activities with multiple communication partners (free play with parents, lunch- or snack-time at home or at school, and speech therapy sessions). Of these children, 19 used multimodal communication, primarily manual signs and speech. Functional word use in both modalities was transcribed. The 50 most frequently used core words accounted for 67.2% of total word use; 16 words comprised core vocabulary, based on commonality. These data are consistent with similar studies related to the core vocabularies of preschoolers and toddlers with typical development, although the number of nouns present on the core vocabulary list was higher for the children in the present study. This finding can be explained by manual sign use of the children with Down syndrome and is reflective of their expressive vocabulary ages.

## Functional word use of children with Down syndrome

Clinicians need to apply a structured and scientific approach to Augmentative and Alternative Communication (AAC) interventions aimed at improving quality of life through achieving the most effective communication possible for individuals who rely on AAC. However, AAC support teams rarely have sufficient knowledge and experience to select vocabulary for functional use for specific contexts, activities, ethnicity, or language groups (Beukelman & Mirenda, 2013). Language sample collection and analysis is an historically used evidence-based practice (Aitchison, 2003) for selecting vocabulary for AAC systems (Kovacs & Hill, 2015). For vocabulary selection, there appears to be a certain set of core words that can be widely used in a range of communicative settings (Banajee, DiCarlo, & Stricklin, 2003). These core words can maximize the potential for spontaneous language generation in spoken, signed, graphic, and/or written (output) modalities. Incorporating core vocabulary into an AAC system may give individuals the opportunity to be engaged in communication and interaction in an appropriate, efficient, and relatively quick manner (Hill, Kovacs, & Shin, 2015; Weighton & Dodd, 2011).



Lee (2001) described a number of features of words characterized as core vocabulary. Such words tend to be (a) the most frequent words in language as a whole; (b) words that are most general, unmarked, or central to the language; (c) the most frequent words in a particular medium; (d) words that are cognitively basic or most salient; (e) the most frequent words for a particular demographic group; (f) words that have the most widespread usage across a wide range of genres, situations, and conversational partners; and (g) words useful for dictionary definitions. In the field of AAC, core vocabulary is most often defined as a small set of approximately 20-50 words for young children and up to 200 - 400 words for adults and used consistently across environments and between communication partners (Baker, Hill, & Devylder, 2000; Chen et al., 2011; Renvall, Nickels, & Davidson, 2013; Weighton & Dodd, 2011; Witkowski & Baker, 2012; Yorkston, Dowden, Honsinger, Marriner, & Smith, 1988). Core vocabulary may account for up to 80% of all words used within communicative contexts. Vocabulary sets described as core vocabulary consist of high-frequency words and represent various parts of natural speech or text (i.e., mostly function words such as pronouns, conjunctions, prepositions, auxiliary verbs, modals, determiners, interjections, and adverbs (Renvall et al., 2013; Witkowski & Baker, 2012). Effective communication requires the combination of both core and fringe vocabulary. Fringe vocabulary items are often large in number, change frequently, and are highly individualized (Yorkston, Honsinger, Dowden, & Marriner,

1989). With these fringe words, which are often content words, an individual is able to reflect on his or her own activities, interests, environments, and personal style (Stuart, Beukelman, & King, 1997).

Spoken language sample studies identifying core vocabulary are available for a variety of groups with no identified disabilities, including toddlers (Banajee et al., 2003), preschool children (Beukelman, Jones, & Rowan, 1989; Fallon, Light, & Paige, 2001; Trembath, Balandin, & Togher, 2007), school-aged children (Robillard, Mayer-Crittenden, Minor-Corriveau, & Bélanger, 2014), adults (Balandin & Iacono, 1999), and elderly adults (Stuart et al., 1997). Core vocabulary has also been identified for children with specific language impairments (Robillard et al., 2014); second-language learners (Boenisch & Soto, 2015); individuals with physical disabilities, some of whom used AAC (Boenisch & Sachse, 2007; Dark & Balandin, 2007; Yorkston, Smith, & Beukelman, 1990); and school-aged children with autism spectrum disorders (Chen et al., 2011). Based on these studies, it can be concluded that the core vocabulary of these populations is highly overlapping. Within the clinical field of AAC, the question has often been raised whether core vocabulary sets are also applicable for children, teens, or adults with intellectual or multiple disabilities.

So far, only three studies have targeted this issue. The first (Mein & O'Connor, 1960), reported spoken core vocabularies of 80 individuals ranging in age from 10 to 30 years, with severe intellectual disabilities and a mental age ranging from 3 to 7 years. Words that were used by more than 50% of the participants were indicated as core vocabulary, leading to a core list of 218 words, which was relatively comparable to the core vocabulary of young children with a comparable developmental age in the 1957 study by Burroughs. The second study, by Chen, Chen, and Chen (2013), explored the spoken core vocabulary of 15 children with intellectual disabilities and their peers without disabilities in fifth and sixth grades during an approximately 30-min dyadic conversation at school. Chronological ages of the children were between 10 – 12 years; IQ scores of the children with intellectual disabilities ranged from 55 to 69. There was 94% overlap for the top 70% most frequently used words in both groups. The third study, by Boenisch (2014), compared the core vocabulary of German children and teenagers (n = 58) with typical development with the core vocabulary used by children with intellectual disabilities of different etiologies. The children with intellectual disabilities were between the ages of 6 and 16 years, with an IQ below 70, attended special schools for children with intellectual disabilities, and were able to use natural speech (n = 44). Language samples were obtained throughout the school day. Boenisch found a very large

overlap between the top 20 (95%), top 50 (94%), and top 100 (87%) words most frequently used by both groups of students. Almost no differences were found between both groups in the frequency of use of different word classes, indicating that the language use of the children and teenagers with intellectual disabilities closely resembles that of peers with typical development.

Van Tilborg and Deckers (2016) reviewed all previously mentioned core vocabulary studies and concluded that core vocabulary, no matter the modality, seems to be comparable for individuals with typical development; monolingual and bilingual individuals with primary language impairments; second language learners; people who rely on text-based AAC; and individuals with physical and/or intellectual disabilities. However, it should be mentioned that many of the studies used different research designs and most analysed language use only in one setting; core vocabulary, as previously defined, is used consistently within and across environments and over a wide range of communication partners. In addition, the few studies of individuals with intellectual disabilities focused mainly on those with mild intellectual disabilities or developmental ages of 4 years and above, and populations with chronological ages of 6 and above. To date, no studies exist that focused on children with intellectual disabilities with chronological ages below school-age and developmental ages below 4 years.



Down syndrome is the most common genetic cause of intellectual disability (Sherman, Allen, Bean, & Freeman, 2007). Owing to its homogeneous etiology, children with the syndrome are considered to be among the most valid reference groups for researching developmental patterns in individuals with intellectual disabilities (van Duijn, Dijkxhoorn, Scholte, & van Berckelaer-Onnes, 2010). These children are usually delayed in learning to speak (Brady, 2008; Kent & Vorperian, 2013) and even after they begin to speak, may be difficult to understand (Roberts et al., 2005). Compared to children with typical development matched on developmental age and based (primarily) on parental reports, Bello, Onofrio, and Caselli (2014) found that children with Down syndrome experience delays in the production of nouns, verbs, and adjectives. Nouns were used significantly more frequently than verbs and adjectives. Other characteristics of spontaneous spoken language of children with Down syndrome include speaking in one-word utterances using nouns or verbs, and limited use of question words, articles, and conjunctions (Abbeduto et al., 2001; Bol & Kuiken, 1989; Chapman & Kay-Raining Bird, 2012). In addition, these individuals may omit grammatical markers and verbs more frequently than peers with the same mean length of utterance (Hesketh & Chapman, 1998).

Gesture production is considered a strength of children with Down syndrome compared to their spoken language skills and also compared to peers with typical development (Galeote, Sebastián, Checa, Rey, & Soto, 2011). In a study by Galeote et al., young children with Down syndrome were found to have comparable vocabulary sizes to those of peers with typical development when the number of gestures was included in vocabulary counts. This shows the importance of accounting for multimodal communication in language studies, particularly in those involving children with Down syndrome. Poor intelligibility and significantly delayed onset of speech are two primary reasons to introduce AAC to children with Down syndrome (Brady, 2008). In Dutch clinical and educational practice, Sign Supported Dutch (SSD) and manual key-word signing are commonly used for this population, as manual sign systems are an important mode of communication, next to or preceding speech (Deckers, Van Zaalen, Mens, Van Balkom & Verhoeven, 2016; Smith & Grove, 1999). The manual signs in Sign Supported Dutch are iconic or arbitrary signs that are deliberately taught by parents, teachers, or speech-language pathologists (Özçaliskan, Adamson, Dimitrova, Bailey, & Schmuck, 2015) and are based on Dutch Sign Language. Manual signs that are iconic in form depict the characteristic actions or attributes of the referent they represent. Different from spontaneous iconic gestures, iconic manual signs are either produced in identical handshape form repeatedly by both the communication partner and the child or by the child alone to refer to the same referent within an observation session. This similarity of form is never observed in spontaneous iconic gestures (Özçaliskan & Goldin-Meadow, 2011), which makes the interpretation of gestures more subjective than when interpreting manual signs.

When manual signs are incorporated in studies of expressive vocabulary development of children with Down syndrome, the vocabulary size is relatively comparable to that of peers with typical development matched for developmental age (Chan & Iacono, 2001; Te Kaat, 2013; Zampini & D'Odorico, 2009). Although several studies show the merits of using manual signs, most studies investigating vocabulary sizes in individuals with Down syndrome only accounted for spoken vocabulary and most often relied only on parental reports, not accounting for the use of other modalities of communication. None of the studies focused on functional use of language in spontaneous language samples in children with Down syndrome. Where noun use seems to be a relative strength in children with Down syndrome, compared to other word classes (Bello et al., 2014), it is not known whether these noun forms are used consistently within environments and between communication partners. The goal of the present study was to determine core vocabulary and the contribution of content and function words to

core vocabulary in young children with Down syndrome with a chronological age between 2 and 7 years, by analyzing (spoken and sign supported) spontaneous language samples in different settings and with several communication partners.

## Method

### Participants

Children with Down syndrome were recruited for this cross-sectional study from specialized Down syndrome polyclinics associated with several hospitals in the Netherlands, through Stichting Downsyndroom, the Dutch association for parents with children with Down syndrome; and speech-language pathologists (SLPs) in clinical practice. Because the Berglund, Eriksson, and Johansson (2001) study showed that some children with Down syndrome do not develop speech by age 5, it was decided to include children with a broader age range. Children were included in the study if they met the following inclusion criteria: (a) developmental age below 48 months, as measured by the Vineland Screener (Van Duijn, Dijkxhoorn, Noens, Scholte, & Van Berckelaer-Onnes (2009); (b) lived in a Dutch-speaking home environment; and (c) had a diagnosis of Down syndrome (trisomy 21). The present study was approved by an institutional review board (NL38926.091.12).

In all, 30 children with Down syndrome participated in the present study: 14 boys and 16 girls. The participants had a mean chronological age of 57 months (*SD* = 16; range: 28–84 months) and a mean developmental age of 28 months (*SD* = 10; range: 12–47 months). Expressive vocabulary ages were determined with the Dutch version of the MacArthur-Bates Communicative Development Inventories: Words and Sentences version (N-CDI), which is recognized as a valuable and valid measure of expressive vocabulary in children with Down syndrome (Deckers et al., 2016). The N-CDI is developed to measure the vocabulary size of children with a chronological or mental age of 16–30 months. Three children reached ceiling levels on the N-CDI. Their expressive vocabulary age was therefore assessed with the Expressive One Word Picture Vocabulary Test (Brownell, 2000). The mean expressive vocabulary age of the children with Down syndrome was 25 months (*SD* = 7; range: 17 – 42 months). The participants in the study lived throughout the Netherlands in different socioeconomic areas. All of the children came from Dutch-speaking households; however, the parents of four of the children had a Turkish background. Of the 30 children, 19 attended schools or daycare centers for mainstream education and 11 attended schools or daycare centers for special education. At the time of the present study, all of the children were attending SLP sessions at least once a week. All of the parents indicated that Sign-Supported Dutch was used at home by both the parents and

the child, and during speech-language therapy sessions by both the SLP and the child. All of the children could speak at least one word, and 17 used manual signs. No picture communication symbols or other AAC devices were used.

## Procedure

As defined in the introduction, core vocabulary comprises a set of words consistently used within and across environments and between communication partners. In the present study, participants' core vocabulary was identified by collecting language samples during interactions in multiple settings and with multiple communication partners during (a) unstructured free play with a parent at home, (b) snack- or lunch-time with a parent at home or with a teacher at school, and (c) speech-language therapy sessions with the SLP. Materials used during free play differed for each child because the children were allowed to play with their own toys, although some common material use was observed (e.g., dress-up games, toy kitchen with equipment, blocks and art). Parents were instructed to (a) play with their child like they normally would, (b) include toys or games according to the child's preference, and (c) allow the child to take the initiative to engage in play with other toys. Snack- or lunch-time activities took place at designated tables, where the children were used to spend their snack- or lunch-time (e.g., at a kitchen table at home, at a table in the school lunchroom). Parents and teachers were instructed to follow the usual mealtime routine. SLPs were instructed to provide therapy as they normally would but to use the recorded therapy session to work specifically on the child's goals related to communication or language in order to evoke sufficient spontaneous language. The language samples taken during these 15- to 20-min interactions were audio and video recorded. Video recording was chosen to identify manual signing used by the children and for visual context when speech intelligibility was decreased.

## Data Transcription and Analysis

For each child, at least the first 10 min of each recorded setting were transcribed. During the transcription process, videotapes were stopped after each utterance and a verbatim transcription was completed of the utterance for both the child and his or her communication partners. Both spoken and signed utterances were transcribed. For every word transcribed, the modality in which that word was produced, either spoken (Sp) or via a manual sign (Si), was added to the transcript. A sample of 100 words per child was constructed, ideally consisting of roughly 33 used words (i.e., 100 words divided by three settings) collected per setting. Of the 30 participating children, 19 had recordings in all three settings. Not all of the children's SLPs participated, which meant that some children took part only

in two settings. The 33-word target was not always reached during spontaneous language use during lunch- or snack-time. This was because mealtime interactions were sometimes constrained by parents who did not want their children to talk while eating (as noted also by Banajee et al., 2003). Therefore, more words uttered during interactions in the other settings had to be included to reach the 100 words threshold for each child.

Transcription rules were based on Trembath et al. (2007) as follows: (a) each utterance was transcribed separately and utterance boundaries were defined by intonation or a pause of longer than 2 s ; (b) when sound repetitions and syllable repetitions occurred, only one whole word was transcribed; (c) fillers or interjections (e.g., *oh/ooh*) and conventional gestures (e.g., waving 'hello' and nodding 'yes' or shaking the head 'no' that are part of a learned, shared, symbolic system) were typed orthographically in a consistent form and counted as words; (d) different forms of a word (e.g., *jump/ jumped*) were transcribed as different words, but combined in the analysis to examine lemma-based words; (e) numbers were typed as nouns; (f) imitated speech, noises, and signs were not included; (g) imitated words, either spoken or in manual signs, were not included; (h) words used in songs and repetitive games were not included; (i) any comments by the children related to recording equipment were omitted from transcription; and (j) names were omitted from transcripts in order to protect confidentiality.

The principles of inter-judge agreement were applied, which can be used to measure reliability of transcripts of utterances of individuals who have limited speech intelligibility (Kovacs & Hill, 2015). Complete inter-judge agreement is obtained by independently transcribing and analyzing a language sample and then resolving discrepancies by consensus as needed (Hegde, 2003). Agreement is usually 100%, unless the raters are unable to reach a consensus (Kovacs & Hill, 2015). Given known problems with both speech and manual sign intelligibility in children with Down syndrome (Kumin, 1994), which can heavily influence the reliability of transcripts, it was chosen to ensure that all language samples were transcribed by two independent raters. Differences that were found in the transcripts, (approximately 2% of the total transcript), were reconsidered by the two raters collectively. In all instances, consensus agreement was reached on appropriate alterations in the transcriptions. About 10% of all utterances were considered unintelligible by both transcribers.

Core vocabulary literature describes several ways to determine which words are core (Van Tilborg & Deckers, 2016). One commonly used metric is to include those words that have an occurrence of at least 0.5 per 1,000 words in a sample (e.g., Balandin & Iacono, 1999; Robillard et al., 2014; Trembath et al., 2007). However, given the relatively small language sample of the present study (i.e.,

3,000 words), applying this threshold would indicate every word used at least twice to be a core word. Instead, core vocabulary was determined in two ways: (a) As the 50 most frequently used words (Chen et al., 2013) in the total sample of 3,000 words, because Boenisch and Soto (2015) showed a significant drop in frequency of word use after the 50$^{th}$ most used words on core vocabulary lists; and (b) On the basis of the principle of commonality. Words were determined as core vocabulary based on commonality, when a particular word was used by 50% or more (i.e., $n \geq 15$) of the children in the total sample (Robillard et al., 2014; Trembath et al., 2007). Applying these two criteria resulted in two word lists with core vocabulary. For both word lists it was determined whether a word was a function (i.e., prepositions, auxiliary verbs, conjunctions, determiners, and pronouns) or a content (i.e., nouns, main verbs, adverbs, adjectives, and negatives) word. Word frequency counts from the transcripts and compilation of both the frequency and commonality list were conducted with R, a platform for statistical computing and graphics (The R foundation, n.d.).

## Results

The total sample consisted of 3,000 words uttered by the 30 participants, of which 420 words were unique. The number of unique words equates to the number of different words used in the transcripts, irrespective of their frequency of use by the children with Down syndrome. Of the total sample, 407 words (i.e., 13.6%) were uttered using a manual sign or via a conventional gesture (e.g., yes, no, bye), 49 of which were unique (i.e., 12%). Of these 49, 32 were nouns. During lunch- or snack-time, 585 words were used, 148 of which were unique (i.e., 25.3%). During sessions with an SLP, 751 words were used, 199 of which were unique (i.e., 26.5%). In the play setting with parents, the children uttered 1664 words, 301 of which were unique (i.e., 18.1%). Language sample recordings to acquire 100 words per child had a mean recording time of 23 min (SD = 6; range: 15.24 min – 36.15 min); mean recording time for lunch- or snacktime was 10 min, for sessions with an SLP 7 min, and for play with parents 6 min. Type-token ratios (i.e., the number of different words divided by the total number of words per setting) were calculated for every child (M = .47, SD = .09).

Table 1 (which includes English translations) shows the core vocabulary list based on the 50 Dutch words that were most frequently used by the children in the present study across all three settings. These 50 words account for 67.2% of total word use, and the 100 most frequently used words account for 80.3% of total word use. Also indicated in the table is whether a word is a content or function word. Of the 50 core words, 27 can be considered function words and

25 as content words. Three words -- hebben (to have), een (a, one), and voor (in front of, for) -- are counted as both function and content words. To have was used as both an auxiliary and a verb. It is important to note that some Dutch words have multiple meanings, such as the word een, which may be the article a or the numerical one; and voor, which may function as an adverb (in front of) or a preposition, as in this is for you (i.e., dit is voor jou). Of the 50 core words listed in Table 1, 22 words were uttered in both spoken and signed modalities, 7 of which were function words and 15 were content words.

Table 2 shows the list of words that achieved a commonality score of at least 15, indicating the words that were used by at least half of the children in the present study. Sixteen unique words were used by at least half of the children, of which only mom and dad are nouns. These core words based on commonality account for 47.1% of the total word use. Ten of these core words based on commonality were uttered in both spoken and signed modalities.

The present study is the first to identify the core vocabulary of young children with Down syndrome (i.e., chronological age between 2;4 (years/months) and 7 years) and a developmental age below 4 years. Developmental ages of the participants were between 1 to 4 years (*M*= 2;4), reflecting a period of early language and communication development. Language samples of 100 words per child were collected across several settings and with different communication partners, resulting in a total language sample of 3,000 spoken and signed words. The 50 most frequently used words accounted for more than 67% of the total sample, indicating that a small set of words, i.e., a core vocabulary, can be identified for young children with Down syndrome. Of the 50 core words, 16 were used by at least half of the participating children. When comparing core words of children in the present study with those of toddlers (Banajee et al., 2003) and preschool children (Beukelman et al. 1989; Fallon et al., 2001; Trembath et al., 2007), it can be concluded that, based on spoken and signed modalities, the core vocabulary of young children with Down syndrome and intellectual disabilities, closely resembles the core vocabulary of young peers with typical development.

Table 1. *The Top 50 Most Frequently Used Words across the Three Settings.*

| Words in Dutch | Translation | Percentage | Words in Dutch | Translation | Percentage |
|---|---|---|---|---|---|
| Ja[a] | Yes | 11.20 | Hallo, Hoi[b] | Hello | 0.67 |
| Nee[a] | No | 7.80 | Ook[a] | Also, as well | 0.67 |
| Die[a] | This, That | 5.10 | Pop(je), Poppen[b] | Doll, puppet | 0.63 |
| Daar[a] | There | 3.83 | Mij, Mijn[a] | My, mine | 0.60 |
| Mama[b] | Mama, mom | 3.47 | Nou, Nu[a] | Now | 0.60 |
| Ik[a] | I | 3.10 | Dit[a] | This | 0.57 |
| Een[a,b] | A, one | 2.00 | Kaas[b] | Cheese | 0.57 |
| Klaar[b] | Ready, finished | 1.67 | Paard[b] | Horse | 0.57 |
| Zo[a] | So | 1.43 | Doen, Doet[a] | To do | 0.53 |
| Jij[a] | You | 1.30 | Koe[b] | Cow | 0.53 |
| Hier[a] | Here | 1.17 | Lekker[b] | Yummy, tasty | 0.53 |
| Nog[a] | More | 1.17 | Meisje[b] | Girl | 0.50 |
| Niet[b] | Not | 1.13 | Dat[a] | That | 0.47 |
| Papa[b] | Papa, dad | 1.10 | Hond[b] | Dog | 0.47 |
| Deze[a] | This, these | 1.03 | Voor[a] | In front of, for | 0.47 |
| Ben, Is, Zijn[a] | To be | 1.00 | Appel[b] | Apple | 0.43 |
| Op[a] | On, all done | 1.00 | Au[a] | Ouch | 0.43 |
| De[a] | The | 0.90 | Drie[b] | Three | 0.43 |
| Kijk(t), Kijken[b] | To look | 0.90 | Geel, Gele[b] | Yellow | 0.43 |
| En[a] | And | 0.77 | Huis[b] | House | 0.43 |
| Weg[b] | Away | 0.77 | Mag[a] | To may | 0.43 |
| In[a] | In | 0.73 | Open[b] | Open | 0.43 |
| Oh/Ooh[a] | Oh | 0.70 | Banaan[b] | Banana | 0.40 |
| Wil/Wilt[b] | To want | 0.70 | Heb, Heeft[a,b] | To have | 0.40 |
| Dag/Doei[b] | Bye | 0.67 | Blauw[b] | Blue | 0.37 |

Note. [a] = function word; [b] = content word.

Table 2. *Core Vocabulary Based on Commonality.*

| Words in Dutch | Translation | N |
|---|---|---|
| Ja[a] | Yes | 30 |
| Die[a] | This, That | 29 |
| Nee[a] | No | 28 |
| Mama[b] | Mama, mom | 26 |
| Daar[a] | There | 24 |
| Ik[a] | I | 22 |
| Zo[a] | So | 22 |
| Een[a,b] | A, one | 21 |
| Klaar[b] | Ready, finished | 20 |
| Hier[a] | Here | 18 |
| Papa[b] | Papa, dad | 18 |
| Niet[b] | Not | 17 |
| Nog[a] | More | 17 |
| Op[a] | On, all done | 16 |
| Deze[a] | This, these | 15 |
| Kijk(t), kijken[b] | To look | 15 |

*Note.* [a] = function word; [b] = content word; *N* = number of children that uttered a particular word.

Recent studies (e.g., Deckers, Van Zaalen, Van Balkom, & Verhoeven, submitted for publication; Polisenka & Kapalková, 2014) showed that the language development of children with Down syndrome, when measured in both spoken and signed modalities, is delayed rather than deviant relative to patterns of typical development. Combined with the results of these recent studies, the findings in the present study may provide additional evidence that the lexical development of children with Down syndrome seems to closely resemble that of children who are developing typically, differing only in the use of multimodal communication.

The core vocabulary of children in the current study serve several syntactic, semantic, and pragmatic functions (see also Banajee et al., 2003; Miller, 1989). Core vocabulary words contained demonstratives (*that, these*), verbs (*to be, to want*), pronouns (*my*), prepositions (*on*), and articles (*the*). Semantic functions included use of agents (*I*), objects (*you*), labeling objects (*that*), actions (*to look*), possession (*my, mine*), affirmation (*yes*), negation (*no*), location (*in*), and termination (*finished, ready*). Pragmatic functions included initiating interaction by attracting attention (*you*), maintaining joint attention (*this, that, these*),

indicating recurrence (*more*), and terminating interaction (*finished, ready*). The types of words in the core vocabulary of participants in the present study appear to be similar in terms of syntax, semantic, and pragmatic functions to those core words identified by previous research with other populations, as mentioned in the introduction. Similarities of the present results to this past research help strengthen the definition of core vocabulary and show the application of core vocabulary across activities, environments, and communication partners, for individuals with typical development as well as children with Down syndrome and intellectual disabilities. Having access to core vocabulary enables young children with intellectual disabilities to meet a variety of syntactic, semantic, and pragmatic functions (Banajee et al., 2003).

As in most clinical core vocabulary studies, the words *yes* and *no* are frequently used by children with Down syndrome. In the current study, these two words alone made up 19% of the total word sample. Investigating the transcripts in closer detail, the overrepresentation of affirmations and negations seems to be due to the fact that communication partners frequently ask yes/no questions of children with Down syndrome. Parents of young children with typical development with language and developmental ages similar to the children in the current study, ask many yes/no questions during interactions with their young child (Eriksson, 2014). This feature is also observed by van Balkom, Verhoeven, Van Weerdenburg, and Stoep (2010) in the communication between children with developmental language delay (i.e., late talkers) and their mothers. These interactions showed lower levels of conversational coherence (i.e., the way both communication partners cooperate to maintain the thread of the conversation), indicating difficulties in fine-tuning and mother's adjusting their language input to the developmental level of their child to establish a coherent conversational context.

Iverson, Longobardi, Spampinato, and Caselli (2006) analysed child-directed language produced by five mothers of children with Down syndrome during a 30 min free-play session. They found that these mothers produced significantly fewer utterances than mothers of younger children with typical development and comparable expressive language skills, and interpreted these results as a maternal simplification strategy to prevent the child from being overloaded with verbal information, which may be the case when asking more closed questions. Zampini, Fasolo, and D'Odorico (2011) found that the frequency of maternal utterances towards children with Down syndrome was influenced by the children's linguistic skills. Mothers tended to talk more when their children's communicative ability was limited, mostly filling in the pauses in their interaction. They gradually reduced their utterance frequency as the child's ability to interact increased, and asked different and more open questions, reflective of a growth in conversational

coherence.

In most core vocabulary studies (see Van Tilborg and Deckers, 2016, for a comprehensive overview) nouns are underrepresented or even absent in core vocabulary lists. In comparison, the core vocabulary list of children with Down syndrome in the current study consisted of slightly more nouns (i.e., 11 out of 50 core words) than core vocabulary lists of peers with typical development. This result may be due to the expressive language ages of the children, with a mean around 2-years of age, and the use of many one-word utterances, in which a noun is more often used by a child to provide content to the utterance. Also, manual signs were counted in the spontaneous language samples in the current study. It is important to note that not every spoken word has a direct translation into a manual sign, especially when it comes to function words such as articles (Bolier, 2010) or manual signs of nouns taught during speech-language therapy or AAC interventions (Adamson, Romski, Deffenbach, & Sevcik, 1992). In the current study, of the 49 unique words in the total language sample uttered via manual signs, 32 were nouns, and 10 of these made it to the core vocabulary list of 50 most frequently used words. In Sign Supported Dutch, manual signs are learned in the immediate everyday context of repeated one-to-one communication with a communication partner. Manual signs are most often used to support speech about an object or action. This creates a highly scaffolded, interactive routing centered on referent-symbol mappings (Özçaliskan et al., 2016). Children with Down syndrome may, therefore, rely on manual signs in particular to convey information about objects and actions in their immediate environment (Dimitrova, Özçaliskan, & Adamson, 2016), explaining the number of signed nouns. The language level of the children and the use of manual signs as an AAC strategy are thus a possible reason for the increased number of nouns on the core vocabulary list in the present study.

A parent or SLP might make a link between a novel word and referent more transparent by calling attention to an object and then stating its name or producing the sound it makes, or following the child's lead and producing labels for objects only after they have entered the child's field of attention (Adamson, Bakeman, & Brandon, 2015). These strategies are more easily related to concrete concepts or content words, such as nouns or nouns used as action verbs (e.g., bike), than to most function words, which often do not have a concrete direct referent in the child's environment. Mothers of children with Down syndrome talk significantly more about objects, using nouns, than mothers of children with language impairments (Kay-Raining Bird & Cleave, 2016). Still, children are able to learn and use more abstract function words, as reflected in the core vocabulary of children with Down syndrome in the current study. In his theory on modeling and vicarious processes,

Bandura (1969) states that individuals acquire words and syntactic structures by exposure to verbalizing models. Some amount of modeling is therefore indispensable for language acquisition. According to this theory, language is learned from models in the environment, and as such, children will have many experiences with and will learn from language used by communication partners in daily life (Tomasello, 2003). Therefore, children encounter core vocabulary used by communication partners from early in their language development. Children are more extensively exposed to specific function words than to specific content words (Segalowitz & Lane, 2000), because these words are more often used by all of the child's communication partners across several activities and settings.

Without any known focus on teaching core vocabulary within speech-language therapy, these core words seem to emerge in the spontaneous interactions of the children with Down syndrome in the current study, either in spoken or in signed modalities. This may not be the case in other children with complex communication needs, who rely on significant others to add core vocabulary to their AAC devices. As stated by Banajee et al. (2003), some words might be difficult to represent visually either through graphic symbols or manual sign, which may result in SLPs not teaching children to use these words during intervention. However, words that are difficult to represent graphically may be taught to young children by modelling the use of these words within activities, as implied by Bandura´s theory of language acquisition. Based on clinical evidence, Van Tatenhove (2009) suggests that, in clinical practice, the balance of core to fringe vocabulary should be at least four to one in AAC systems or on language activity cards. For example, a child with Down syndrome who relies on AAC is engaged in a sandpit activity in a classroom. The child's AAC system is a communication board with, following the examples of Van Tatenhove (2009), up to 50 core vocabulary words. Attached along the top of this core vocabulary board is a spiral bound row of 20 strips, each displaying 10 specific activity, place, topic, or partner-specific, fringe vocabulary words. In one of the rows, sandpit vocabulary (i.e., fringe vocabulary), such as sand, water, scoop, castle, and bucket, is accessible. Once the activity is finished, the fringe vocabulary can be exchanged with fringe vocabulary for a subsequent activity. The core vocabulary on the communication board never changes and, following the findings of the present study and the larger research base in core vocabulary, would be the same for other children in the classroom as well. The fringe vocabulary should be individualized to the person´s needs, wants, and communication environment. Vocabulary, core and fringe combined, must reflect children's changing and evolving communication needs and contexts, in order to allow for successful interactions in a variety of contexts and maintain sensitivity to the child's current

and future developmental language abilities and skills (Marvin, Beukelman, & Bilyeu, 1994). The core vocabulary list for young children with Down syndrome who rely on AAC should at least include the 16 core words based on commonality found in the present study.

## Limitations

Three possible limitations of the present study require consideration. First the sample size is relatively low, at just 30 participants. Although small sample sizes are common in core vocabulary studies and the recruitment of participants with Down syndrome is not easy, this restriction may limit the generalizability of the findings of this study. In small sample sizes, wide variability in performance, as reflected in the range of both developmental age and expressive vocabulary scores in the present study, may limit conclusions about development of children with Down syndrome as a group (Patterson, Rapsey & Glue, 2013). However, the wide variability between children is found to be common in the vocabulary development of children with Down syndrome (Galeote et al., 2008), indicating that our research group may closely resemble the characteristics of the Down syndrome population.

A second limitation is the relatively small language samples collected -- 100 words per child -- although influential studies such as Banajee et al. (2003) collected only 150 words per child. Given the delayed vocabulary development and increased degree of unintelligible speech common in young children with Down syndrome (Brady, 2008), collecting and transcribing reliable language samples is time consuming. Thus, in order to include the same number of words in the total sample for every child, the 100-word threshold was chosen. The children with higher expressive vocabulary scores reached this threshold after about 15 min of recording, while others did not reach threshold until 35 min or more. Nevertheless, given that the present study is the first to focus on children with intellectual disabilities and developmental ages below 4 years, the results provide important insights into core vocabulary and functional vocabulary use in this participant group. In addition, the results are highly comparable with core vocabulary lists of studies in other populations. Future studies should investigate the core vocabulary of this group in more detail, including participants whose intellectual disability is not related to Down syndrome.

A final limitation is the uneven distribution of words uttered in the three different settings. Although language samples in different settings and with different communication partners were collected, about 50% of the total sample of words were uttered in the play setting with parents and under 20% of the words were uttered in the lunch- or snack-time. Lunch- or snack-time settings may

evoke fewer utterances among children because they are discouraged from talking and eating at the same time. Future studies of core vocabulary in children with intellectual disabilities should consider other contexts as well (e.g., pre-school classrooms, activities with siblings or peers) in order to identify commonality of vocabulary use over a range of contexts and communication partners. Nonetheless, given the definition of core vocabulary as a small set of words, used consistently across environments and communication partners, and that changes little over time (Baker et., 2000; Chen et al., 2011; Renvall et al., 2013; Weighton & Dodd, 2011; Witkowski & Baker, 2012; Yorkston et al., 1988), it can be hypothesised that results will be comparable to the results of the present study.

## Conclusion

The goal of the present study was to determine core vocabulary in children with Down syndrome and a developmental age below 4 years, by analyzing spontaneous language samples in different settings and with several communication partners. The 50 most frequently used core words accounted for 67.2% of total word use; 16 words were determined to be core vocabulary based on a commonality criterion (i.e., a word was used by at least half of the participants). Words in the core vocabulary of young children with Down syndrome appear to be similar in syntactic semantic, and pragmatic functions to core words identified by research into other populations, although the contribution of content words to the core vocabulary of the children with Down syndrome seems higher than in other groups. As reflected in the present study, AAC use and vocabulary selection during AAC intervention, such as the overrepresentation of noun use in Sign Supported Dutch, influences functional word use and determination of core vocabulary lists. A higher number of content words, relative to function words, may also reflect the expressive vocabulary age of the children with Down syndrome in the present study. The results of the present study strengthen the definition and applicability of core vocabulary as a construct for the field of AAC. To achieve the most effective communication possible, vocabulary selection in AAC should best be based on the emerging evidence relating to functional language use of children with intellectual disabilities.

# Chapter 4

## Predictors of lexical development in children with Down syndrome

This chapter is based on Deckers, S. R. J. M., Van Zaalen, Y., Van Balkom, H., & Verhoeven, L. (submitted). Predictors of receptive and expressive vocabulary development in children with Down syndrome. Revision submitted to International Journal of Speech Language Pathology.

## Abstract

There is a lack of longitudinal data on predictors of vocabulary development in children with Down Syndrome. In typically developing children, many internal and external predictors of vocabulary development have been determined before. The purpose of the present study was to investigate the role of these variables in the receptive and expressive vocabulary development of children with Down syndrome. The present study used a longitudinal design in young children with Down syndrome to study the vocabulary development over a period of 1;6 years and investigate the possible predictive role of child-related and environmental variables. Receptive vocabulary development was best predicted by the adaptive level of functioning, and early receptive vocabulary skills. Expressive vocabulary development was best predicted by the adaptive level of functioning, receptive vocabulary, maternal educational level, level of communicative intent of the child, attention skills and phonological/phonemic awareness. Predictors found in these children resemble those predicting vocabulary development in typically developing children, as shown in other studies. These results add to the research base showing that lexical development in children with Down syndrome seems to be delayed rather than deviant from typical development.

# Predictors of lexical development in children with Down syndrome

Vocabulary can be defined as all the words that are known (i.e. receptive vocabulary) and used (i.e. expressive vocabulary) by an individual, and this doesn't only include spoken words. A broad range of child- and environment-related predictors of vocabulary development have been described in the literature about young typically developing children. To date, researchers in the field of Down syndrome (DS) show a one-sided focus on only a few of these predictors, namely early vocabulary levels, nonverbal mental abilities, short-term memory skills and chronological age. The present study used a longitudinal design to investigate the role of all these child- and environment-related variables in the receptive and expressive vocabulary development of children with DS.



DS is characterized by developmental delays in all areas of function, although the degree can vary among individuals (Rogers, Gordon, Schanzenbacher, & Case-Smith, 2001). Where children with Down syndrome show a congruence between receptive vocabulary skills and nonverbal cognitive skills (Chapman, Schwartz, & Kay-Raining Bird, 1991; Rice, Warren, & Betz, 2005; Rosin et al., 1988), they are typically delayed in learning to speak (Brady, 2008). According to Abbeduto, Warren and Conners (2007), the transitioning from the pre-linguistic stage into intentional symbolic communication of either words or signs is delayed in children with DS, not appearing until 18-36 months or later. Children with DS produce their first word around the average age of 1;9 years (Stoel-Gammon, 2001), though substantial individual variation exists (Berglund, Eriksson & Johansson, 2001). For instance, Gillham (1990) found that it is not until the age of 3;9 years that some children with DS uttered their first words, while Berglund et al., (2001) noted that some children started to talk at or after age five.

Once they begin to produce words, their progress relative to their typically developing peers is very slow (Brady et al., 2004; Yoder & Warren, 2004). Typically developing children reach the 50-word milestone around the age of 18 months, which is often indicative of a vocabulary spurt and the start of combining words to two-word utterances (Berglund et al., 2001). Berglund et al. , however, found the 50-word milestone was only reached by about 25% of children with DS at age three, by about 50% at age four and about 75% at age five. About 10% to 20% of the children with DS had less than 10 words in their spoken vocabularies when they were already three to 5 years old. Spoken vocabulary of children with DS at ages 3;0 and 4;0 was comparable with that of typically developing children at ages 1;4 and 1;8 respectively. Ninety percent of

three year-olds and 94% of five year-olds with DS is able to speak one or more words (Martin, Klusek, Estigarribia, & Roberts, 2009).

Subsequent growth in early expressive vocabulary is slow compared with expectations for typically developing children (Berglund et al., 2001). Most studies of vocabulary development in children with DS, however, have an important limitation. The production of words is often taken into account only in its vocal modality (Berglund et al. 2001), whereas many children with DS spontaneously produce numerous gestures and signs (Galeote et al., 2008; Galeote et al., 2011). Galeote et al. (2008) studied the relationship between cognitive development and vocabulary size in both the vocal and gestural modality in 66 Spanish children with DS. Children with DS were found to have comparable vocabulary sizes as typically developing peers when gestures were also taken into account. Between the ages of one and 5;6 years 87% of 330 children with Down syndrome used speech and manual signs simultaneously (Berglund et al., 2001). It is recognized that interventions using Augmentative and Alternative Communication (AAC) strategies may help children with DS communicate earlier and more successfully. A poor intelligibility of speech and the significantly delayed onset of spoken words are two primary reasons to introduce the use of manual signs to children with DS (Brady, 2008). An example of an AAC intervention strategy applying manual signing in clinical practice is key-word signing (Windsor & Fristoe, 1991), which mirrors spoken language in a motor-visual mode, accompanying and supporting the speech.

In Dutch clinical and educational practice, where the present study was conducted, Sign Supported Dutch (SSD), fingerspelling and key-word signing, are commonly used in young children with DS, mostly as Simplified (manual) Sign System (Bonvillian, 2002), where other forms of AAC are often neglected in Dutch practice. As a result these children use manual sign systems as an important mode of communication (Deckers, Van Zaalen, Mens, Van Balkom & Verhoeven, 2016a). A large number of young children with DS is ready to communicate through a motor-visual (manual) representation of language by ten to twelve months of age (Kumin, 2003). The use of gestures and manual signs requires less advanced neuromuscular execution skills than speech. In gesturing and signing, the sender of the signal also receives bimodal feedback, that is proprioceptive as well as visual information on produced hand and arm movements (Tomasello, 2008). These combined features of manual signs and speech facilitates communication development. Wright, Kaiser, Reikowsky and Roberts (2013) showed that a naturalistic manual sign intervention including gestures, manual signs and spoken words appeared to be more effective on expressive vocabulary skills in children with DS than interventions that taught spoken words alone.

By the age of five years, most children with DS have usually outgrown the need to use signs as their primary communication system due to an increased intelligibility of their speech, unless they have a significant hearing loss or verbal apraxia or dysarthria, and the use will gradually fade out over the next year(s) (Kumin, 2003). The signs used in SSD stem from Sign Language of the Netherlands, and are therefore actual signs and not just (deictic or representational) gestures. Assessment of vocabulary size in these children with DS should therefore also incorporate their manual sign vocabulary size (Deckers et al., 2016a). There is agreement in the literature that there are large individual differences in vocabulary development among children with DS (Galeote et al., 2008). How these huge differences in the development of vocabulary can be explained remains unclear until now.

In typically developing children aged 2 – 5 years, early receptive vocabulary skills tend to be a strong predictor of later receptive and expressive vocabulary skills (Chiat & Roy, 2008; Watt, Wetherby, & Shumway, 2006). The variation in this receptive vocabulary development itself in typically developing children aged 4 – 5 years can be explained by several predictors, such as gender, ethnic background, child's temperament, hearing status, phonological and phonemic awareness, being read aloud to (i.e. parent-child book reading), maternal education, language spoken in the home environment, family structure, sibship size, and socioeconomic status (see Taylor, Christensen, Lawrence, Mitrou, & Zubrick, 2013 for a comprehensive overview). Expressive vocabulary development in typically developing children aged 1-6 years was found to be predicted by the level of communicative intent, attention distractibility and joint attention (Brady, Marquis, Fleming, McLean, 2004; Farrant & Zubrick, 2011; Sally, Panneton, & Colombo, 2013). In preschool children aged 3-5 years, working memory capacity also showed a strong impact on initial vocabulary levels, but also on vocabulary growth (Ebert et al., 2013). However, differences in working memory capacity become less important when vocabulary size increases (Gathercole, Willis, Emslie, & Baddeley, 1992). Finally, an interactionist perspective to language and vocabulary learning stresses the importance of the child's early environment and social interactions (Snow, 1994). An important variable that should thus be accounted for is the number of communication partners within the social network (Blackstone & Hunt-Berg, 2003), next to the family's Social Economic Status (SES). Following the model of holistic development (Fröhlich & Haupt, 2004), the vocabulary development of children is interrelated with several other developmental domains, such as cognition, memory, attention, adaptive level of functioning and socio-emotional skills. Based on recent neurocognitive studies (see Denes, 2011), and theoretical models describing the interdependencies amongst these developmental domains

and vocabulary, language and communication development (Jackendoff, 2002; Van Balkom, 2009), we proposed an adapted model showing the underlying processes beneath language learning and communication performance (see Deckers, Van Zaalen, Stoep, Van Balkom, & Verhoeven, 2016b). All predictors of vocabulary development described above can be related to that adapted model.

Given that word learning in children with DS seems to be delayed rather than deviant from typical development (Polisenka, & Kapalková, 2014), the same predictors that play a role in typical development may also play this role in the development of receptive and expressive vocabulary in children with DS. Receptive vocabulary is regarded as a strength as described in the DS phenotype (Abbeduto et al., 2003), and is therefore often only taken into account as possible predictor for expressive vocabulary development. In children with DS, several variables have already been associated with expressive vocabulary development, such as chronological age (Chapman, Hesketh, & Kistler, 2002) and nonverbal mental age (Miller et al., 1993). In a study of children and adolescents with DS aged 5 to 20 years, Chapman, Seung, Schwartz and Kay-Raining Bird (2000) found the contribution of chronological and nonverbal mental age appeared to be mediated through receptive vocabulary skills. Furthermore, poor concentration, distractibility and poor inhibitory control are well-documented risk factors related to language and vocabulary problems of children with DS (Cornish & Wilding, 2010). Cornish et al. (2012) indeed found lower receptive vocabulary levels to be associated with early problems in inattention and hyperactivity. Finally, medical issues should be accounted for when investigating predictors of vocabulary development. Visootsak, Hess, Bakeman and Adamson (2013) found that a congenital heart defect, present in about half of children with Down syndrome, accounted for a significant part of variation in language delay even when it is repaired. This implicates that also medical conditions should be accounted for when studying predictors of vocabulary development.

Most of the vocabulary studies in children with DS used a cross-sectional design and only focussed on a limited number of predictors. Only a few studies have used longitudinal designs to assess the vocabulary development of children with Down syndrome, due to which there is a lack of data on trends in vocabulary development (Zampini & D'Odorico, 2013). Insofar longitudinal studies on vocabulary development have been conducted only a limited number of predictors was incorporated (e.g. Oliver & Buckley, 1994; Miller, 1999; Berglund et al., 2003). Zampini and d'Odorico (2013) did use a multivariable longitudinal design to study expressive vocabulary development and its predictors in 18 children with DS between two and four years of age. They found significant prediction of

chronological and mental age and earlier expressive vocabulary scores. However, these predictors were also the only variables investigated.

Even more recently, Næss, Lervag, Lyster and Hulme (2015) followed the receptive and expressive vocabulary development of a cohort of 43 6-year-old children with DS. Although the longitudinal design and the inclusion of a control group of 57 3-year-old typically developing children matched on nonverbal mental abilities are strengths of their study, it also has a major weakness. Measures included in the study to predict vocabulary development were only verbal short-term memory and early vocabulary levels, not looking at many of other possible predictors. Næss et al. (2015) concluded that later vocabulary skills could not be significantly predicted by both early vocabulary measures and verbal short-term memory. A possible explanation for this finding is that Næss et al. used a composite score combining the results of expressive and receptive vocabulary tests into one latent variable. Since receptive and expressive vocabulary may have different predictors, constructing this latent variable may have compromised their findings. Despite the often mentioned deficit in verbal short-term memory in children with DS, vocabulary learning skills are not as impaired as might be expected (Laws, 1998; Mosse & Jarrold, 2011; Næss, Lyster, Hulme and Melby-Lervag, 2011). There seems to be no evidence for a longitudinal predictive relationship between verbal short-term memory skills and later vocabulary knowledge in children with DS between 4 and 8 years old (Melby-Lervag et al., 2012; Næss et al., 2015). To date, researchers in the field of DS show a one-sided focus on only a few predictors of vocabulary development in DS, namely early vocabulary levels, nonverbal mental abilities, short-term memory skills and chronological age. Several predictors mentioned in typically developing children have not been taken into account before.

In a cross-sectional, multiple case-study, Deckers et al. (2016b) showed how to use the International Classification of Functioning-Child and Youth (ICF-CY) in classifying the myriad of contributing factors to communication and receptive and expressive vocabulary in young children with DS, in order to identify personal and environmental facilitators and barriers. The goal of the present study is to use a longitudinal design to investigate the early receptive and expressive vocabulary development over a period of 1;6 years in children with DS, taking into account the predictive role of child- and environment-related predictors as found in typically developing children. The research question of the present study was as follows: Which child- and environment-related factors are predictive of the receptive and expressive vocabulary development in children with DS?

# Method

## Participants

Children with DS and their parents were recruited from special Down Syndrome teams in several hospitals in the Netherlands, via speech language pathologists in clinical practice and through an advertisement on Facebook and in the magazine of 'Stichting Downsyndroom', referring to the Dutch association for parents with a child with Down syndrome. Inclusion criteria for the children where: (a) chronological age between 2;0 and 7;6 years of age, and (b) having Dutch as a natural home language environment. Studies investigating predictors of vocabulary development in typically developing children, as described in the introduction, focussed on children between 1 – 6 years old. Because the Berglund et al. (2001) study showed only few children with DS started to speak at age 1 and some children with DS had not started speech development at age five, we decided to include children with a broader age range in order to include a research population that was early in their vocabulary development. An exclusion criterion was the presence of severe auditory or visual disabilities. Interested parents were sent an information letter and brochure, explaining the nature and the purpose of the study. The letter was accompanied by an application form and a questionnaire containing questions about child characteristics, such as the presence of a comorbid diagnosis. The form and the questionnaire could be returned to the authors in a stamped addressed envelope. Application forms and questionnaires were returned by 36 parents. All children had been genetically diagnosed with Trisomy-21. The mean age of these 36 children was 4;5 years (SD = 1;5 years; range: 2;1 – 7;0 years) and 20 were boys. All children came from Dutch-speaking households, however, the parents of two children had a Turkish background, and lived all across the Netherlands. One child was known with a double diagnosis of Autism Spectrum Disorder. The study is approved by an Institutional Review Board, CMO Arnhem-Nijmegen (NL38926.091.12).

## Materials and Procedure

The data acquisition focussed on functioning in several areas related to vocabulary development. First, parents were sent multiple questionnaires pertaining to these areas. The Dutch version of the MacArthur-Bates Communicative Development Inventories (N-CDIs; Zink & Lejaegere, 2002) was used for receptive and expressive vocabulary. CDI's were developed for spoken vocabulary. However, given that in the Berglund et al. (2001) study 87% of children with DS used manual signs and the extensive use of SSD in Dutch clinical practice, parents could also indicate a word as 'produced' by their child when they used a manual

sign for a particular word. The concurrent and predictive validity of the adapted N-CDI for both spoken and signed words in children with DS was found to be high by comparing results from the parental report with utterances produced by children during 15 minutes interactions with parents and speech language therapists and with scores on expressive vocabulary tests (Deckers et al, 2016a). A word was seen as produced or understood irrespective of the modality. A word that could both be signed and spoken by a child was counted as one word to determine vocabulary size. Other questionnaires used in the present study were (a) Vineland Screener for the adaptive level of functioning (Van Duijn, Dijkxhoorn, Noens, Scholte, & Van Berckelaer-Onnes, 2009) which is reflective of the developmental age of a child, (b) Subscale Working Memory from the Behaviour Rating Inventory of Executive functions Preschool version (BRIEF; Huizinga & Smidts, 2012), (c) Child Behaviour Checklist 1½ - 5 (CBCL; Achenbach & Rescorla, 2000) for behavioural and emotional problems, attention distractibility and temperament, (d) The Bridge: Emergent literacy skills (Pierce, Summer & O'DeKirk, 2005) for insight in book reading experiences and phonological/ phonemic awareness, and (e) Social Networks Questionnaire (Blackstone & Hunt-Berg, 2003) for the number of communication partners. Furthermore, parents were sent a questionnaire including questions about socioeconomic status (i.e., family income and educational level of parents) and other child- and environmental related variables such as chronological age of the child, siblingship size, and educational level or involvement of the child. Parents returned filled out questionnaires via mail.

Second, parents, speech language pathologists (SLP) and other professionals were asked to send the medical and/or institute dossiers of the children. These dossiers were checked for relevant test scores for hearing or vision status.

Third, several (standardized) tests were administered. The tests used in the present study were (a) Dutch version of the Receptive One-Word Picture Vocabulary Test (ROWPVT; Brownell, 2000) for receptive vocabulary, (b) Auditory discrimination task (Verhoeven & Vermeer, 2001), (c) Auditory working memory test (Gathercole, Pickering, Hall & Peaker, 2001), and (d) Communicatieve Intentie Onderzoek (CIO; Van der Meulen, Slofstra, & Lutje-Spelberg, 2013) for the assessment of communicative intent. The CIO also includes an observation of joint attention and parental support and responsiveness. More information about the parental questionnaires and (standardized) tests can be found in Appendix A. Child data were collected by trained research assistants (i.e. fourth year speech language pathology students) unaware of the research question and goals. Research assistants were trained in both the procedure, using signs and in scoring of test instruments. For example, the ROWPVT was

administered with both the use of spoken and signed words (e.g. 'Where is the car [sign of car]?'. All assessments were videotaped and it was verified whether the research assistants correctly followed the procedures. Scoring of the assessments was provided by the research assistants and verified by the first author, a clinically certified educational and psychological scientist, according to the instruction manuals of the tests. Agreement between both raters was found in 96% of the scores.

Given the (a) extensiveness of our data sampling, (b) to minimize the intrusiveness of participation for the child and his/her parents, and (c) the often found slow development of vocabulary in children with DS, two waves of data collection were chosen with a time frame of 1;6 years (T1 and T2).

## Data analysis

First, we constructed a latent variable for receptive vocabulary based on the scores of both the parental questionnaire, the N-CDI, and the ROWPVT (for a comprehensive overview of using latent variables for child language, see Bornstein & Putnick, 2012). Both measurements have their own strengths and weaknesses and the best insight in the child's capabilities is gained with a combination of assessment methods (Deckers et al., 2016a). In standardized tests, language is provoked in an isolated situation and may not represent active use of these words for a specific child during the day. The attention span of young children is an important factor influencing the assessment and therefore the result is likely to prove unrepresentative of the child's abilities (Feldman et al., 2005). Also, the level of receptive vocabulary is often harder to indicate reliably by parents than the expressive vocabulary, since the latter is more directly observable. Therefore, a combination of both scores was used in the present study as a measure for receptive vocabulary. Composite scores were calculated with the use of SPSS for this latent variable for receptive vocabulary.

Second, descriptive statistics were calculated for the number of words children could express according to the N-CDIs. We calculated this separately for the number of words that children could speak or sign and for the total number of different words that were indicated by parents at the N-CDI. In order to investigate whether children with DS significantly develop their expressive vocabulary, related-samples Wilcoxon Signed Rank Tests ($\alpha = .05$) were conducted for either growth in total vocabulary size, the number of spoken words and the number of signed words. To test whether there were differences in vocabulary levels between boys and girls and for educational setting of the child, Mann-Whitney U-tests were conducted ($\alpha = .05$).

Third, given the relative broad range in chronological age between participants in the present study, partial correlations ($\alpha = .05$) were conducted to correct for the influence of chronological age and the related years of experience with language input and communicative environments. Correlations were conducted at T1 and T2 for all variables and receptive and expressive vocabulary measures at that time-point. Partial correlations were also conducted to test whether predictor variables at T1 correlated significantly with vocabulary measures at T2. In these analyses, raw scores were used. According to Riccio, Sullivan and Cohen (2010), raw scores may be more sensitive than norm-based scores to smaller changes in functioning over time and are therefore most useful in measuring growth within individual students. Among children with disabilities, it may be especially important to assess change via raw scores, given that these children are likely to develop at a slower rate than their typically developing peers. Using standard scores would then prevent from detecting development, since these standard scores may even deteriorate when development is slower than typical development (Sullivan, Winter, Sass, & Svenkerud, 2014). Also, age-equivalent (AE) scores may not be useful in measuring growth in children with development disabilities. The growth seen in raw scores over time may not be large enough to be seen with AE scores and therefore the use of AEs may mask (small) individual changes in ability levels over time (Sullivan et al., 2014).



Fourth, for all predictor variables and vocabulary measures growth scores were calculated (i.e. score at T2 minus score at T1), since we were also interested in variables that correlated with growth in vocabulary scores. Partial correlations were conducted between growth in expressive and receptive vocabulary scores and scores at T1 and growth of all predictor variables.

Fifth, all predictor variables that showed significant correlations with vocabulary measures were entered stepwise in linear regressions at T1, T2 and for growth in vocabulary scores ($\alpha = .05$). Stepwise regression is helpful in the identification of important predictors if a lot of candidate predictors in the model are involved of which is not clear how they will affect the model (Dugard, Todman, & Staines, 2010), as was the case in the present study. Beforehand, z-scores were calculated for all measurements and used in the regressions. Based on the successive regression models, we constructed a structural relations model as a summary of these results.

## Results

The mean age of the 36 children participating at T1 was 4;5 years (SD = 1;5 years; range: 2;1 – 7;0 years) and 20 were boys. The mean mental age corresponding to the adaptive level of functioning was 1;9 years (SD = 0;8 years; range: 0;10 – 3;4 years). All parents and children participating at T1 were invited to participate at T2 as well. Of all children, 20 were included at T2. Drop outs were due to (a) medical issues with children occurring around T2 (n = 2), (b) family issues (n = 3), (c) parents not returning filled in questionnaires, although tests were already administered (n = 4), and (d) parents indicated they no longer wanted to participate, but did not provide an explanation (n = 7). The mean age of the participating children at T2 was 5;10 years (SD = 1;5 years; range: 3;2 – 7;2 years). The mean mental age corresponding to the adaptive level of functioning at T2 was 2;8 years (SD = 0;11; range: 1;4 – 4;2 years).

A wide range of parent reported vocabulary sizes was evident from the returned N-CDIs. The total expressive vocabulary size of the children with DS according to 36 parents ranged from 7 to 699 words with a mean of 202 words at Time 1, and ranged from 7 to 702 words with a mean of 316 words at Time 2. The mean scores, standard deviations, and ranges for N-CDI scores for expressive vocabulary are reported in Table I. Of the 36 parents that responded at T1, 20 also filled out the N-CDI at T2. At T1, one child showed ceiling levels, due to which it was not included at T2. At T2 two children showed ceiling effects at the N-CDI, with parent reported expressive vocabulary sizes around 700 words. Wilcoxon Signed Rank tests showed significant growth in the total expressive vocabulary size ($Z = 168$; $p < .01$) and in the total number of words spoken ($Z = 114$; $p < .01$), but not in the total number of words signed. Mann-Whitney U-tests were conducted to test whether gender differences or differences between children in mainstream (n = 25 at T1) or special education (n = 11 at T1) existed. No significant differences were found between boys and girls for receptive and expressive vocabulary levels at T1 and T2. Significant differences were found for the number of spoken words at T1, but not at T2 ($U = 10.5$; $p = .01$) and in receptive vocabulary composite scores at T1 ($U = 11.0$; $p = .02$), favouring children in mainstream education over children in special education. No significant differences in growth of vocabulary were found between boys and girls and for educational setting.

Secondly, partial correlations were conducted at T1 and T2, controlling for chronological age for all variables and receptive and expressive vocabulary measures at that time-point. Partial correlations were also conducted to test whether predictor variables at T1 correlated significantly with vocabulary measures at T2. All significant correlations for receptive, spoken and total (i.e.

Table 1. *Descriptive Statistics of Number of Words Expressed at T1 and T2.*

| | N | Mean | Median | SD | Range |
|---|---|---|---|---|---|
| **T1** | | | | | |
| Total expressive vocabulary size | 36 | 202.0 | 173.0 | 162.9 | 7 – 699 |
| Total # of words spoken | 36 | 140.9 | 137.0 | 185.8 | 0 – 699 |
| Total # of words signed | 36 | 84.9 | 57.0 | 83.8 | 0 – 301 |
| **T2** | | | | | |
| Total expressive vocabulary size | 20 | 316.3 | 387.0 | 198.4 | 7 – 702 |
| Total # of words spoken | 20 | 329.7 | 381.0 | 213.9 | 0 – 702 |
| Total # of words signed | 20 | 78.9 | 44.0 | 116.0 | 0 – 351 |
| **Growth** | | | | | |
| Total expressive vocabulary size | 20 | 314.3 | 97.0 | 141.4 | -15 – 373 |
| Total # of words spoken | 20 | 188.8 | 118.0 | 147.8 | -12 – 403 |
| Total # of words signed | 20 | -6.0 | -26.0 | 135.7 | -301 – 338 |

Note. The total vocabulary size is not the sum of # of words spoken and signed. It is based on the number of different words expressed by the child, irrespective of the modality. Many children used both modalities for the expression of one specific word.



both spoken and signed) vocabulary are displayed in Table 2. No significant correlations were found between predictor variables and the size of sign-only vocabulary. Developmental age, the level of communicative intent, auditory discrimination skills, phonological and phonemic awareness, foundations of reading, alphabet knowledge as well as the level of withdrawn behaviour show significant correlations with receptive and expressive vocabulary measures at both T1 and T2. Most of these factors at T1 also show significant correlations with receptive and expressive vocabulary at T2. Maternal educational level shows a significant positive correlation with expressive vocabulary at T1 and the number of communication partners in the direct family at T1 correlates significantly with the number of spoken words at T2. Receptive and expressive vocabulary measures are highly correlated at all time-points.

Next, partial correlations were conducted between growth scores of receptive and expressive vocabulary with predictor variables at T1 and growth in scores on these predictor variables, controlling for the effect of chronological age. No significant correlations were found for growth in receptive vocabulary scores and for growth in total vocabulary scores. A significant correlation was found between growth in spoken vocabulary and growth in attention skills ($r = .68$; $p < .01$),

Table 2. *Significant Partial Correlations between Receptive and Expressive Vocabulary and Predictors at T1 and T2, Corrected for the Influence of Chronological Age.*

| | Receptive vocabulary T1 | Receptive vocabulary T2 | Spoken expressive vocabulary T1 | Spoken expressive vocabulary T2 | Total expressive vocabulary T1 | Total expressive vocabulary T2 |
|---|---|---|---|---|---|---|
| **Predictors at T1** | | | | | | |
| Developmental age (VS) | .84** | .84** | .75** | .72** | .76** | .80** |
| Communicative intent (CIO) | .76** | .83** | .40* | .76** | .47** | .78** |
| Auditory discrimination (AD) | .74** | - | .47** | - | .50** | - |
| Spoken Expressive vocabulary (N-CDI) | .60** | - | ***** | .84** | .91** | .78** |
| Total Expressive vocabulary (N-CDI) | .64** | - | .91** | .84** | ***** | .84** |
| Phonological/phonemic awareness (Bridge) | .58* | - | .62** | .81** | .71** | .80** |
| Alphabet knowledge (Bridge) | .56* | - | - | - | .48* | - |
| Foundations of reading (Bridge) | .54* | .73** | - | - | .48* | .61* |
| Receptive vocabulary (RV) | ***** | .79** | .60** | .69** | .64** | .75** |
| Withdrawn (CBCL) | - | -.57* | -.51* | -.74** | -.55* | -.83** |
| Maternal education | - | - | .47* | - | .56** | - |
| Communication partners in direct family (SN) | - | - | - | .54* | - | - |
| **Predictors at T2** | | | | | | |
| Alphabet knowledge (Bridge) | | .89** | | .72** | | .78** |
| Developmental age (VS) | | .80** | | .57* | | .66* |
| Spoken Expressive vocabulary (N-CDI) | | .65** | | ***** | | .98** |
| Total Expressive vocabulary (N-CDI) | | .71** | | .98** | | ***** |
| Foundations of reading (Bridge) | | .68** | | .72** | | .67** |
| Communicative intent (CIO) | | .61* | | - | | - |
| Phonological/phonemic awareness (Bridge) | | - | | .61* | | .53* |
| Receptive vocabulary (RV) | | ***** | | .65** | | .71** |
| Withdrawn (CBCL) | | - | | - | | -.61* |

*Note.* * $p < .05$; ** $p < .01$

indicating that children who increased in attention skills over time developed their spoken vocabulary more.

All predictor variables that showed significant correlations with receptive and expressive vocabulary measures (see Table 2) were entered stepwise in multiple-linear regressions at T1, T2 and for growth in vocabulary scores. A significant regression equation was found for all dependent variables, indicating that receptive and expressive vocabulary were found to be predicted by at least one predictor variable. Model summaries and explained variances by the models are displayed in Table III. Explained variance of the models (adjusted R2), which is indicative for the model quality, varies between 33% for growth in spoken expressive vocabulary to 83% for total expressive vocabulary at T2.

Table 3. *Model Summaries and Descriptives of Multiple Linear Regressions.*

| Dependent variables | F | df1 | df2 | Adjusted $R^2$ | p |
|---|---|---|---|---|---|
| Receptive vocabulary T1 | 108.94 | 1 | 31 | .77 | <.001 |
| Receptive vocabulary T2 (predictors at T1) | 42.02 | 1 | 17 | .70 | <.001 |
| Receptive vocabulary T2 (predictors at T2) | 28.23 | 1 | 16 | .62 | <.001 |
| Spoken expressive vocabulary T1 | 56.51 | 1 | 33 | .62 | <.001 |
| Spoken expressive vocabulary T2 (predictors at T1) | 20.46 | 2 | 11 | .75 | <.001 |
| Spoken expressive vocabulary T2 (predictors at T2) | 12.89 | 1 | 14 | .44 | <.01 |
| Total expressive vocabulary T1 | 22.72 | 2 | 19 | .67 | <.001 |
| Total expressive vocabulary T2 (predictors at T1) | 15.17 | 1 | 17 | .44 | <.01 |
| Total expressive vocabulary T2 (predictors at T2) | 35.88 | 2 | 12 | .83 | <.001 |
| Growth in spoken expressive vocabulary | 6.53 | 1 | 10 | .33 | .03 |

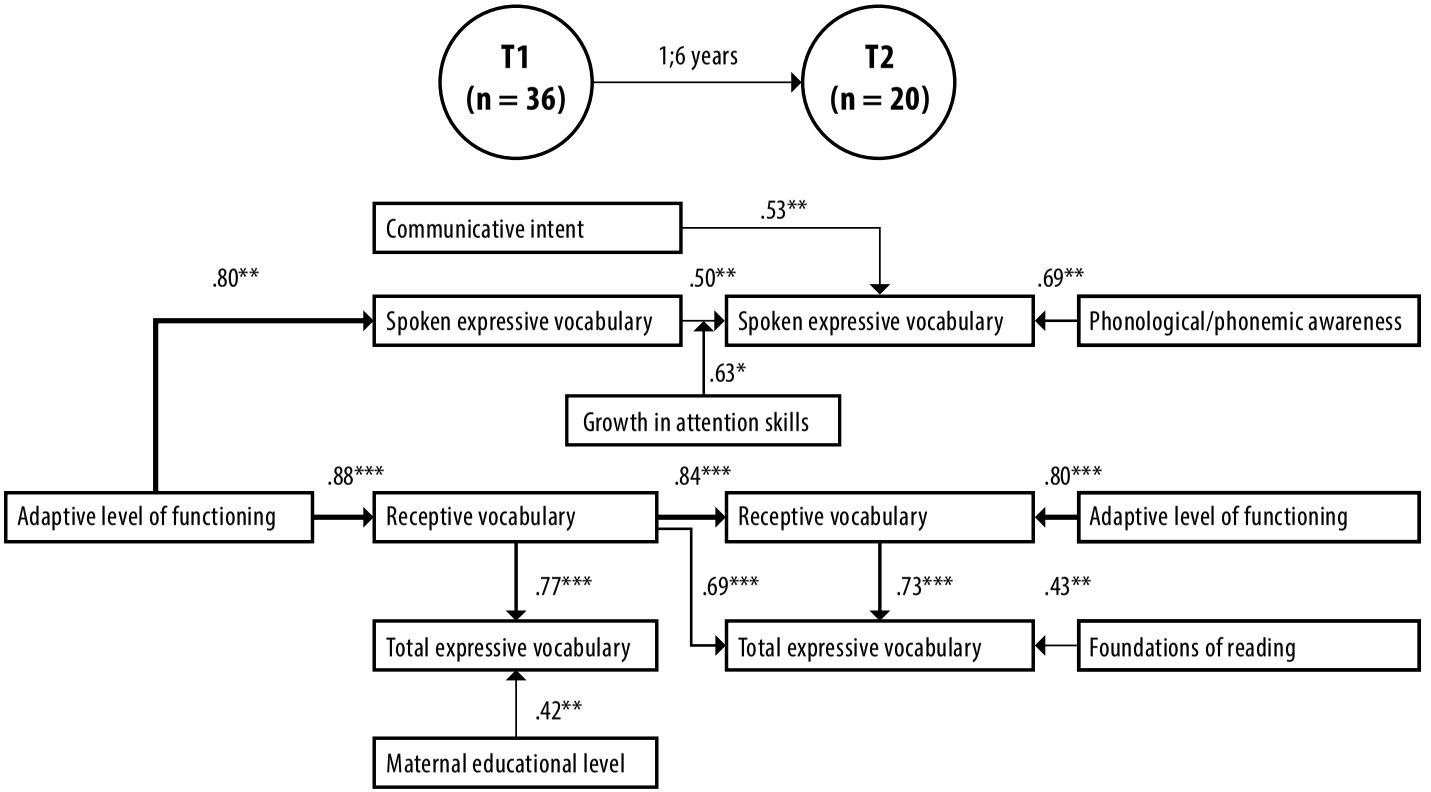
Predictors from each of these equations are modelled in Figure I. The figure presents a structural relations model of receptive and expressive vocabulary development based on the successive stepwise multiple linear regressions. Bèta weights and significance levels are displayed in the figure. Early levels of receptive vocabulary could best be explained by the adaptive level of functioning, as is also the case for early spoken expressive vocabulary. Later levels of receptive vocabulary could best be predicted by early receptive vocabulary levels and the adaptive level of functioning. Early total expressive vocabulary (i.e. both spoken and signed words) was found to be explained by receptive vocabulary and the maternal educational level. Later spoken expressive vocabulary in children with DS was mostly influenced by phonological/phonemic awareness, early levels of communicative intent and

early spoken vocabulary. The growth in spoken expressive vocabulary itself could best be explained by the growth in attention skills. Later total expressive vocabulary could best be predicted by receptive vocabulary levels, both early and later, and by skills related to foundations of reading.

## Discussion

The aim of the present study was to investigate the early receptive and expressive vocabulary development in young children with DS and to identify the possible role of predictors, as identified in typical development, over a period of 1;6 years. Several studies showed that supporting communication through AAC techniques such as signs has many benefits for individuals with DS and does not inhibit or discourage verbal speech development or production (Millar, Light, & Schlosser, 2006), as is also shown in the present study. Many of the young children with DS in the present study used signs at the start of the study. The children, as a group, showed a significant growth in receptive and spoken expressive vocabulary, but not in signed vocabulary.

A wide range in growth is seen in the 20 children with DS who participated in the longitudinal study. Some children verbally expressed almost 400 more words after the period of 1;6 years. Other children verbally expressed only a few more words or even verbally expressed less words. The range in use of signs is even wider. Children used about 300 signs more at T2 and other children, especially the older ones, used about 300 signs less at T2. Our results closely resemble the findings by Kumin (2003) and Vandereet, Maes, Lembrechts and Zink (2011), that by the age of five years, children with intellectual disabilities, among which children with DS, have usually outgrown the need to use signs as their primary communication system. Kumin interprets this may be due to an increased intelligibility of their speech. As suggested by Capone (2007), if a child's meaning representation is intact, but poorly linked to his/her phonological encoding, the child may express a concept more readily in gestures or signs. In fact, gesture and sign production may help create a more precise and concrete image linked to the word (Bello, Onofrio, & Caselli, 2014), since it helps focusing their attention on the object or referent that must be named and allow them to create a link between the perceived object or action and its spoken label (Butterworth, 2003; Goldin-Meadow, 2007). Stefanini, Caselli and Volterra (2007) state that the productive deficit of children with DS is not due to the representation of meaning, but is merely due to the linking of meaning to speech production, which may be affected by problems in phonological skills, working memory, and speech motor execution (Kumin, 2006). They found




*Note.* * $p < .05$; ** $p \leq .01$; *** $p \leq .001$

Figure 1. Structural relations model of receptive and expressive vocabulary development based on successive stepwise multilinear regressions.

that children with DS, in comparison with mental age matched peers, produced significantly more iconic gestures that were semantically related to the meaning of pictures of a vocabulary test and thus conveyed the correct information lacking in speech. There is a high degree of imprecision in the physical execution of signs and gestures by children with DS (Stefanini et al, 2007), so intelligibility might be a problem in using signs as well. However, imprecisely signed words may be easier to comprehend by communication partners than imprecisely articulated speech, evoking more (and different) responses by communication partners. It is recommended to investigate this in future research.

The present study found several important predictors for early (T1) and later (T2) vocabulary sizes in children with DS, both internal and external factors to the child. Predictors of vocabulary development in children with DS found in the present study will be discussed in the order according to the strength of the relation as reported in the structural relations model in Figure I: (a) the children's adaptive level of functioning predicting both receptive and spoken expressive vocabulary, (b) the children's receptive vocabulary predicting total expressive vocabulary, (c) the maternal educational level predicting early total expressive vocabulary, (d) the children's early levels of communicative intent predicting later spoken expressive vocabulary, (e) the children's phonological and phonemic awareness predicting later spoken expressive vocabulary, and (f) the children's growth in attention skills predicting growth in spoken expressive vocabulary. All six aspects will be defined and discussed in the next paragraphs.

## Adaptive level of functioning

The adaptive level of functioning can be defined as the collection of conceptual, social and practical skills that have been learned by individuals in order to function in their everyday live (American Association on Intellectual and Developmental Disabilities, 2008), and is reflective of the child's level of participation in daily activities in relevant contexts (Case-Smith, 2001). Skills related to the adaptive level of functioning in children with DS are found to be acquired in a similar sequence and according to a similar trajectory as typically developing peers (Dolva, Coster, & Lilja, 2004; Van Duijn et al., 2010). Van Duijn et al. (2010) used the Vineland Screener to determine the level of adaptive functioning of 984 Dutch children with DS, aged between 0 and 12 years, in comparison with the level in typically developing peers. The Screener distinguishes four components of adaptive functioning: communication, daily living skills, socialization and motor and mobility skills.

In our study, the level of adaptive functioning predicted both receptive and expressive vocabulary. In typically developing children of 3 to 6 years old

(Muluk, Bayoglu, & Anlar, 2013), and in young children with developmental disabilities resulting from neurological aetiologies (DeVeney, Hoffman, & Cress, 2012), daily living skills, socialization and motor and mobility skills significantly predicted receptive and expressive language and vocabulary skills, suggestive of the global nature of development in young children, which may also be the case in children with DS. Relatively strong skills in daily living skills and socialization, but relative weaknesses in developing motor and communication skills in children with DS are often found (Dykens, Hodapp, & Evans, 2006; Van Duijn et al., 2010). Being able to be independent in taking care of basic every day needs is important and increases the child's sense of competence and self-esteem (Sperber & Wilson, 1986). Practical independence often leads to more social inclusion and participation in preschool, school and in the community for children with DS (Buckley & Bird, 2009).



Poor fine and gross motor skills may be affecting performance of complex self-care tasks (Dolva et al., 2004), possibly limiting the opportunities of a child to interact and manipulate with objects, which influences concept building, and the intelligibility of manual signs. Being able to move and/or take an active role in activities increases opportunities for social interaction and word learning (Sacks & Buckley, 2003). Fine motor skills have also been related to play abilities and to manipulate toys. McCathren et al. (1999) reported that children's level of play was significantly related to later expressive vocabulary, providing them with experiences with several concepts. This relates closely to the theory of embodied cognition, where the body plays a significant causal role in cognitive processing. A word like "cup" automatically activates motor information and memory, since the language concept is closely linked to the motor action or experience (Borghi & Cimatti, 2010).

For children with DS, socialization is usually a strength. They are very able to pick up feelings and behaviours, although they may not understand the spoken language accompanying these signals (Buckley, Bird, & Sacks, 2002). Children with more developed social skills will elicit more responses from communication partners, having more experiences with language contexts, learning more different words. The ability to use these words to communicate increases a child's ability to manage social situations, to ask for what they want and to explain how they feel. A growth in receptive vocabulary skills allows parents to explain actions and to reason with their child (Buckley et al., 2002). This means that vocabulary development and the adaptive level of functioning are interrelated, and growth in skills on the one domain affects functioning in the others and vice versa.

## Receptive vocabulary

In typically developing toddlers it is often found that early performance in language areas predicts later language performance and that early receptive vocabulary skills tend to be a strong predictor of both receptive and expressive performance (Watt, Wetherby, & Shumway, 2006; Chiat & Roy, 2008), as was shown in this study with children with DS as well for receptive and total expressive vocabulary. Chapman (2006) and Roberts, Price and Malkin (2007) state that the level of receptive vocabulary is partly a result of life experience, closely related to chronological age, and cognitive development, closely related to the adaptive level of functioning. Comprehension in children with DS is coherent with more general cognitive abilities in the early years (Bello et al., 2014). Where Chapman et al. (2000) found the receptive vocabulary to be the most important predictor for spoken expressive vocabulary in children with DS, we did not find this direct relationship. An explanation for our finding can be seen in the study by Nash and Snowling (2008) who found that a lower verbal productivity in children with DS, as compared to typically developing peers, was unlikely to be due to impoverished lexical content or lower receptive skills, given that the children were matched on receptive vocabulary knowledge. Rather, Nash and Snowling concluded that the verbal productivity might be more affected by having poorly organized word knowledge, or is due to problems with phonological representations.

## Phonological and phonemic awareness

Children who have phonological awareness are able to identify and manipulate units of oral language, such as words, syllables and onsets and rhymes. Phonemic awareness refers to the specific ability to focus on and manipulate phonemes (i.e. individual sounds) in spoken words. The results of the present study show significant partial correlations between receptive vocabulary and phonological/phonemic awareness in children with DS. This may point towards a possible indirect influence from receptive to spoken expressive vocabulary, which is mediated by phonological awareness. As individuals' receptive vocabulary increases, their phonological awareness develops via a process of lexical restructuring (Bowey, 2001). According to the lexical restructuring hypothesis, vocabulary can provide the foundations for phonological sensitivity, or the combination of both phonological and phonemic awareness skills (Dickinson, McCabe, Anastasopoulos, Peisner-Feinberg, & Poe, 2003), as was shown in a study in children with intellectual disabilities of several aetiologies between the age of six and nine years old (Van Tilborg, Segers, Van Balkom & Verhoeven, 2014). When the receptive vocabulary of a child grows, children will become more easily aware of shared rhymes or sounds between words, since they will learn multiple

words that start (or end) with the same sounds or that rhyme, making it also easier to distinguish between sounds and words in expressive vocabulary. A stronger language foundation, by an increased receptive vocabulary, may facilitate the acquisition of alphabet and sound knowledge (Van Bysterveldt, Gillon, & Foster-Cohen, 2010). In the early stages, vocabulary development may thus predict later phonological and phonemic awareness skills. In children and adolescents with DS, it was earlier found that receptive vocabulary skills predicted performance on both phonological decoding tasks and word identification measures (Kay-Raining Bird, Cleave, & McConnell, 2000).

The present study in children with DS showed that phonological/phonemic awareness was a significant predictor of spoken vocabulary. Spoken word production requires the retrieval of phonological information. As a consequence of increased receptive vocabulary knowledge, phonological representations become more precisely specified and distinct, since representing lexical entries in terms of a limited set of common phonemes is more efficient than representing the phonological structure of each word separately (Metsala & Walley, 1998). Access to a variety of phonological representations, which is enhanced by a growing receptive vocabulary, enables the child to produce a diversity of sounds and words, increasing its spoken expressive vocabulary. Although phonological awareness increases, intelligibility of speech may not be enhanced directly or to a great extent, given that fine speech motor control may not be established (Kumin, 2003) or phonological encoding is not automatised yet. Accordingly Van Zaalen-op ‘t Hof (2009), if, due to a lack of automatisation, much attention is needed to complete phonological encoding, fewer resources remain for other processes, particularly articulation and monitoring of speech. Consequently, a high level of language production difficulties remain and intelligibility is only slightly increased. The speech disorder associated with DS is therefore not a simple delay caused by an intellectual disability, but, next to the motor problems, rather a disorder of phonological acquisition (Dodd & Thompson, 2001; Roberts et al., 2005). These findings show that it is important for future studies to differentiate between spoken and signed words when investigating vocabulary development in children with DS, as other predictors may account for development.



## Maternal educational level

In 3- and 4-year-old typically developing children, mother’s educational level was positively correlated with their child’s abilities to choose between four pictures, to define words, to know the use of an object, to count, and to name antonyms (Muluk et al., 2013). Mothers with higher social economic status backgrounds used longer utterances and more diverse vocabulary when talking

to their two-year-old typically developing children, which was associated with larger subsequent child vocabulary growth (Hoff, 2003). The results of the present study show that maternal educational level relates to vocabulary skills in children with DS as well, although we have not investigated the language input and used vocabulary of mothers directly. Other studies have shown that the language input of mothers of children with DS closely resembles the input of mothers of typically developing children. For example, Zampini, Fasolo and D'Odorico (2011) found that mothers of children with DS produced similar number of utterances per minute to mothers of typically developing children at the same level of linguistic skill. More recently, Kay-Raining Bird and Cleave (2015) compared mothers' talk about familiar and unfamiliar nouns and verbs to children with DS, language impairments or typical development. They found that mothers of children with DS talked significantly more about familiar nouns than other mothers, providing additional semantic information and talking more about physical or distinguishing features of the nouns. Kay-Raining Bird and Cleave conclude their study provides evidence that mothers of children with DS, as equally to the other mothers, "fine-tune their talk to their young children in ways that would support novel noun learning" (p. 25). Provided these results, we hypothesise that mothers of children with DS with a higher educational level are even more able to fine-tune their talk to their child, supporting vocabulary learning. Our results indicate that this effect of maternal educational level seems to fade out later in vocabulary development. Where the educational level is no longer found to be a predictor of later vocabulary skills in the present study, the level of communicative intent and attention skills of the child become more important. Zampini et al. (2011) explained this by their finding that mothers of children with DS tend to talk more when their children's communicative ability is limited, stimulating conversation and filling pauses, gradually reducing their frequency of utterances as the child's communicative intent increases. The early levels of communicative intent were found to predict later spoken expressive vocabulary for the children with DS.

## Communicative intent

An intentional communicative act can be defined as a persuasive act or expression to reach a goal through another person (Van der Meulen et al., 2013), either by use of gestures, vocalizations, signs or spoken words in order to convey a message (Wetherby & Prizant, 1993). As we also found in our study in young children with DS, the level of communicative intent was already found to predict later receptive and expressive vocabulary skills in typically developing toddlers (Wetherby et al., 2002), in premature children (Ulvund & Smith, 1996) and in

young children with developmental disabilities of different aetiologies (Brady et al., 2004; DeVeney et al., 2012). Children who exhibit higher rates of intentional communication develop language earlier and show increased vocabulary growth than their peers who exhibit low rates (Sarimski, 2002), as is reflected in the results of children with DS in the present study. These higher rates of communicative acts result in more opportunities for communication partners to respond and shape communication development (MacCathren, 2000). Yoder, Warren, Kim, and Gazdag (1994) found that mothers provided more verbal modelling when children communicated intentionally. More frequently verbal modelling, and thus providing more parental input, leads to better expressive vocabulary skills in children by providing them more opportunities for active participation (Brady et al., 2013).



## Attention skills

Farrant and Zubrick (2011) found that the effect of maternal education and warm parenting on vocabulary development in one-year olds was completely mediated by attention skills and parent-child book reading when the children were three years old. A finding which is also seen in the present study in children with DS. The growth in spoken expressive vocabulary was found to be significantly predicted by the growth in attention skills. Increased attention demands are found to negatively affect aspects of auditory comprehension (Murray, Holland, & Beeson, 1997) and spoken vocabulary (Hula, McNeil & Sung, 2007) in typically developing individuals. Furthermore, attention is a necessary and sufficient condition for working and long-term memory storage and retrieval (for an overview of studies, see Kurland, 2011). Children are more likely to learn words for objects if they are able to focus their attention on the object. Once the child's attention is given to the object, caregivers may more easily provide referential input. This increases the likelihood of the child associating the object with the spoken or signed word and using the spoken or signed word in communicative settings. Children who have greater regulation of attention, may be more likely to experience more opportunities for vocabulary learning when they attend to their parents' speech or signs (Salley, Panneton, & Colombo, 2013). Moore, Oates, Hobson, and Goodwin (2002) observed that infants with DS showed significantly smaller durations of looking and a slower latency and longer looks away from a play setting than typically developing infants. The mothers of infants with DS seemed to play a more pivotal role in directing their infants' attention during play than mothers of typically developing peers. Moore and colleagues concluded that a more forceful maternal style during interactions served to maintain or obtain attention in their infants with DS. Mothers of children with DS seem to provide

natural compensation strategies to obtain and maintain the child's attention, when the child is not yet able to focus its attention by himself.

## Limitations and implications

Three important limitations of the present study will be discussed. The sample size is relatively low, with 39 children with DS at T1 and 20 children at T2. Although the recruitment of participants with DS is not easy and small sample sizes are common in longitudinal studies with children with DS, this restriction limits the generalisability of the findings of this study. In small sample sizes wide variability in performance, as seen in the wide range of vocabulary scores in the present study, may limit conclusions about typical development in children with DS (Patterson, Rapsey & Glue, 2013). These differences are, however, found to be common in their vocabulary development (Galeote et al., 2008). Based on successive stepwise multilinear regressions, we constructed a structural relations model. The total model could not be assessed on goodness-of-fit in the present study, due to a lack of power given the small sample size and the differing N between T1 (N = 36) and T2 (N = 20). As such, the total model should be interpreted with caution. In a future study, our structural relations model of vocabulary development should be tested in a larger population with a Structural Equation Modeling (SEM) technique, both in children with DS as well as in a control group of typically developing children. SEM is a statistical approach to model means and covariances among multivariate data and allows for a simultaneous analysis of all variables in the model, visualized by a graphical path diagram. The present study does provide important insights in predictor variables of receptive and expressive vocabulary development of children with DS. Given that it is the first study to include a wide range of child- and environment-related factors in predicting development, results need confirmation in order to investigate the predictive role of all variables at once in both children with DS as well as typically developing children.

A second limitation may be that we used many parental questionnaires for several variables. There are some studies that questioned the accuracy of parental questionnaires, particularly in atypical groups such as DS, in which parents tend to over- or underestimate the abilities of their child (Roberts et al., 1998). However, we used instruments that have proven to be reliable or valid in children with DS, such as the Vineland Screener (Van Duijn et al., 2009) and the N-CDI (Deckers et al., 2016). Furthermore, it has often been stated that children with DS appear to have an 'on-demand problem', due to which skills demonstrated in spontaneous settings may be absent during assessment of that skill (Fidler &

Nadler, 2007), indicating that results on standardized tests would not likely have resulted in more reliable measurements of variables included in the present study. Another limitation of the present study could be that we have not investigated motor skills in depth as indicated earlier. Advanced motor skills provide infants with more opportunities for experiencing the world (Iverson, 2010). Changes in posture, locomotion, and object-manipulation and being able to move through their environments makes children to experience objects in their environment in a new manner, possibly influencing how children develop their vocabulary (Behrens & Hauch, 2011). Children with slow motor development were found to have smaller vocabularies (Viholainen et al., 2002). Future studies would benefit from investigating the role of fine, gross and speech motor skills in vocabulary development in children with DS in more detail.

Given the broad range of the number of manual signs used by the children with DS in the present study, following Galeote et al. (2008), it is beneficial to explain to parents that sign instruction in the early stages of language and vocabulary development helps to improve initial communication of children with DS, and increases speech production (Millar, Light, & Schlosser, 2006; Schlosser & Wendt, 2008). It is also beneficial for parents and SLPs to keep using signs after the moment a child with DS starts to speak (Vandereet et al., 2011), for two reasons. The first reason for continuing the use of signs augmenting speech is that the visual-motor modality of signs may increase understanding of a message by the child, especially when auditory discrimination or auditory working memory are weak or impaired. Secondly, it is a clinical observation during the 1;6 years of the present study that parents of children who started to speak more, left out using signs themselves as well. Possibly as a result, we observed that some children used less signs overall, even for words they did not produce verbally, leading to a decrease in number of words expressed or a stagnation in vocabulary development. This trend should be investigated in more detail in future studies. Thirdly, the use of manual signs slows down the speed of speech production and shows an increase in intonation, prosody in the language input of the caregivers, which facilitates the speech and information processing in children with DS (Beukelman & Mirenda, 2013).

## Conclusion

Within a longitudinal design, the present study showed several predictors for receptive and expressive vocabulary development in young children with DS. Predictors found in these children seem to resemble those also predicting vocabulary development in typically developing children, as found in other studies. Receptive vocabulary development was best predicted by the adaptive level of functioning, and early receptive vocabulary skills. Expressive vocabulary development was best predicted by the adaptive level of functioning, receptive vocabulary, maternal educational level, level of communicative intent of the child, attention skills and phonological/phonemic awareness. These results show that it is important for both the research and clinical field to focus on both child- and environment-related variables when looking at vocabulary development in children with DS. Following the model of holistic development (Fröhlich & Haupt, 2004) as described in the introduction, the vocabulary development of children with DS is interrelated with several other developmental domains, which should be accounted for in both research and clinical practice.

# Appendix A

Table A. *Overview and Description of Used Measurements.*

| **Measurement** | Variable | **Description** |
|---|---|---|
| The Dutch version of the Communicative Development Inventories (N-CDI) | *Receptive and expressive vocabulary* | The N-CDI (Zink & Lejaegere, 2002) is a parental questionnaire for expressive and receptive vocabulary. The Words and Sentences form of the N-CDI (for children with a chronological or mental ageof 16–30 months) contains a checklist of 702 words divided into several categories. We used an adapted version in that parents could indicate for each single word whether their child used spoken language, signs or both. The CDI and its adapted version have been found to be valid and reliable measurements in children with Down syndrome (Deckers et al., 2016; Galeote et al., 2016). |
| Receptive One-Word Picture Vocabulary Test (ROWPVT) | *Receptive vocabulary* | The ROWPVT (Brownell, 2000) measures the receptive vocabulary and is an individually administered, norm-referenced assessment of how well persons age 2 years 0 months to over 80 years can match a word that is heard to objects, actions, or concepts presented in full-color pictures in a multiple-choice format (i.e., "Where is the…?". The ROWPVT consists of 190 items. Instructions were adapted specifically for children with Down syndrome and included signs as well (f.e., 'Where is the car? {makes manual sign of car}) |
| Vineland Screener (VS) | *Adaptive level of functioning* | The VS is a norm-referenced assessment and consists of 90 items adhering to four components of adaptive functioning: communication, daily living skills, socialization and motor skills. The reliability and validity of the Dutch version of the Vineland Screener were found to be high in typically developing children (Van Duijn et al., 2009) and in a study of 984 children with Down syndrome aged 0 – 12 years (Van Duijn et al., 2010). |
| The Bridge | *Emergent literacy skills* | The Bridge: Emergent literacy (Pierce, Summer & O'DeKirk, 2005) is a portfolio rating scale of oral language and emergent literacy behaviours. The Bridge has 11, six-point rating scales that address: (1) foundations of reading (book knowledge/ appreciation/print awareness/story comprehension); (2) foundations of writing (motor and cognitive/linguistic skills and understandings); (3) alphabet knowledge; (4) phonological/phonemic awareness; and (5) oral language understandings in literacy interactions. Early analysis of the psychometric properties by Pierce et al. suggested that interrater reliability ranged from .60-.85 for all children; however, interrater reliability was 100% for children with intellectual and multiple disabilities. |

| | | |
|---|---|---|
| Behaviour Rating Inventory of Executive Function (BRIEF) | *Working Memory* | The subscale working memory from the BRIEF (Huizinga & Smidts, 2012) contains 10 items. The BRIEF is a validated and norm-referenced assessment and measures the range of executive function in (pre)school-aged children. The subscale working memory measures the ability to maintain information in the focus of one's attention to complete a task or generate the appropriate response. Although no validity or reliability measures of the BRIEF in children with Down syndrome exist, the instrument shows great potential for describing the nature of executive function difficulties in children with Down syndrome (Lee et al., 2011). |
| Auditory Memory Test | *Auditory working memory capacity* | The Auditory Memory Test (Gathercole, Pickering, Hall & Peaker, 2001) measures the longest list of items that a person can remember. The child hears a list of words, ranging from one to six successive words, and has to recall the string of words. There were six items for each string length. An adapted version was used which did not require speech production abilities (Peeters, Verhoeven, De Moor, & Van Balkom, 2009).In this version, the child heard a string of words and after two seconds the child heard another string of words (the same string or a slightly different string) (e.g., [boat], [knife], [cap], versus [boat], [window], [cap]). The child had to decide whether the two successive strings of words were identical or not. The internal consistency of this task in the study of Peeters et al. in young children with intellectual and/or physical disabilities was very high with Cronbach's alpha ranging from .95 to .97. |
| Auditory Discrimination Test | *Auditory discrimination* | To assess auditory perception abilities, the Auditory Discrimination Task of the standardized Dutch Language Proficiency Test was administrated (Verhoeven & Vermeer, 2001). In this task, the child was presented with minimally differing word pairs and had to indicate whether the words in a pair sounded alike. Peeters et al. (2008) constructed an adaptation for children with intellectual and/or physical disabilities. For each minimally differing word pair two color drawings were presented of concrete one-syllable words that were related to each other in sound, but differed minimally in either their first- or last consonant, or in their vowel (e.g. bel [bell] and bal [ball], or bijl [axe] and pijl [arrow]). The child was asked to point or indicate the word that was mentioned. The task had 20 items and the reliability of the task was found to be high in children with intellectual and/or physical disabilities (Peeters et al., 2009; Van Tilborg et al., 2014). |

| | | |
|---|---|---|
| Communicatieve Intentie Onderzoek [Communicative Intent Assessment] (CIO) | *Communicative intent, joint attention and parental support and responsiveness* | The CIO (Van der Meulen, Slofstra-Bremer, & Lutje Spelberg, 2013) is a validated and norm-referenced instrument to observe the preverbal and early stages of the verbal development and joint attention skills within a play context with a parent and subsequent with an SLP. The instrument also scores the competences and limitations in communication performance of communication partners during play/conversation with the child. |
| Child Behaviour Checklist (CBCL) | *Behavioural and emotional problems, attention distractibility and temperament* | The CBCL is a validated and norm-referenced parental questionnaire containing 99 items and identifying specific behavioural, emotional, and social problems that characterize preschool and school-aged children (Achenbach & Rescorla, 2000). Parents are asked to rate their child for how true, on a 3-point Likert scale, each item is now or within the past 6 months. Eisenhower, Baker and Blacher (2005) report good internal consistencies for all CBCL scales in preschool children with intellectual disabilities, some diagnosed with Down syndrome. |
| Social Networks Inventory | *Number of communication partners* | The Social Networks Inventory (Blackstone & Hunt-Berg, 2003) is an interview instrument to identify existing relationships of an individual and to identify opportunities and barriers to participation in life activities. The Inventory is specifically developed for the field of Augmentative and Alternative Communication and individuals with communication and language disabilities. Communication partners and their barriers and facilitators for communication with a child can be identified in five circles: (1) direct family, (2) friends, (3) classmates and acquaintances, (4) paid professionals, (5) partners in the outside world. |
| Dossiers | *Hearing and vision status* | Dossiers of all children were collected and checked for reports of hearing and vision screenings. |

# Chapter 5

## A comprehensive assessment of lexical development in Down syndrome: integrative profile of communication performance

This chapter is based on Deckers, S. R. J. M., Van Zaalen, Y., Stoep, J., Van Balkom, H., & Verhoeven, L. (2016). Communication performance of children with Down syndrome: An ICF-CY based multiple case study. Child Language Teaching and Therapy, 32, 293 – 311.

## Abstract

Enhancing communication performance skills may help children with Down syndrome to expand their opportunities for participation in daily life and to support lexical development. It is a clinical challenge for speech-language pathologists to disentangle various mechanisms that contribute to the language and communication problems that children with Down syndrome encounter. Without clarity of different levels of functioning, appropriate interventions may be poorly conceived or improperly implemented. In the present study, the International Classification of Functioning, Disability and Health –Children and Youth Version (ICF-CY) framework was used to classify contributing factors to communication performance in a multiple case study of six young children with Down syndrome. Within a comprehensive assessment, we identified individual and environmental facilitators and barriers, leading to an integrative profile of communication performance (IPCP) for each child. Whereas these six children shared a developmental, and/or level of lexical development and/or level of communicative intent, the children faced similar but also unique personal and environmental factors that play an important role in their communication performance and related lexical development. Our data reveal that a combination of different factors may lead to the same language outcomes and vice versa, based on a unique pattern of interdependency of ICF-CY domains. Planning interventions for enhancing communication performance and lexical development in children with Down syndrome should therefore be based on a comprehensive view on the competences and limitations of every individual child and its significant communication partners. This evaluation should address facilitators and barriers in body functions, structures, activities, participation and environment, with a specific focus on individual strengths. The ICF-CY provides a useful framework for constructing an integrative profile that serves this purpose.

# A comprehensive assessment of lexical development in Down syndrome: Integrative profile of communication performance

Communication performance can be defined as a person transmitting (expressive communication) and receiving verbal and non-verbal messages (receptive communication) in different environments (World Health Organization, 2007). Communication enables a person to express wants and needs, to socialize with others, to convey information, and to participate in broader society (McLeod and Threats, 2008) when intended to transmit information (Wetherby et al., 1997). An intentional communicative act is defined as a persuasive act or expression to reach a goal through another person (Van der Meulen et al., 2013). For children with Down syndrome, the ability to effectively convey communicative intent is often restricted due to their delayed lexical development and problems with intelligibility (Guralnick et al., 2011; Raghavendra et al., 2012).

A clinical challenge for speech-language pathologists (SLPs), teachers, and behavioural and educational scientists is to recognize and distinguish the role of several developmental domains that influence communication performance and lexical development in the individual child with Down syndrome. Children with similar assessment results, or with the same diagnosis, may actually perform very differently from each other in and with varying success in functional situations in the natural environment (WHO, 2001: 4). This is why Light and McNaughton (2015) stated that there is an urgent need to extend assessment in the field of communication performance to plan interventions for lexical development:

*(a) to build on the individual's strengths and focus on the integration of skills to maximize communication, (b) to focus on the individual's participation in real-world contexts, (c) to address psychosocial factors to maximize the resources that the individual brings to bear on the communication process, and (d) to focus on environmental factors to eliminate opportunity barriers and maximize social supports for the individual with complex communication needs. (p. 9)*

The individual and environmental strengths may serve as latent enablers or facilitators for the child and its significant social partners (Lee, 2011b; Roberts et al., 2007; Rowland et al., 2012), because these strengths may link to a cascade of interrelated intervention techniques and methods (Beukelman and Mirenda, 2013; McCauley and Fey, 2006).

The International Classification of Functioning, Disability and Health – Children and Youth Version (ICF-CY) asserts that disability is a complex social phenomenon that involves the interaction between the intrinsic characteristics of the individual and the extrinsic factors associated with the social context in which the person lives (WHO, 2007). Within the field of SLP, the ICF-CY is already broadly applied to classify and discuss communicative disabilities in children (Simeonsson, 2003), with typical speech acquisition (McLeod, 2007), with speech impairments (McLeod, 2006; McLeod and McCormack, 2007), with language impairments (Washington, 2007; Westby, 2007), and with fluency disorders (Yaruss, 2007). Using the ICF-CY code system, data on a child's functioning can be arranged, described, summarized and exchanged in a standardized way for multidisciplinary teams. In the ICF-CY perspective, functioning encompasses how children use their individual resources (i.e. individual strengths and environmental facilitators) and how involved they are in contexts where they usually spend time (i.e. the level of participation; WHO, 2007). A universal view of human functioning advocates respect for individual differences in ability (Campbell and Skarakis-Doyle, 2007). Individual differences are variations in strengths between individuals, but also dissimilarities in strengths within an individual, Revealed in different social contexts. Strikingly, in using the ICF-CY framework in clinical practice, these individual differences in ability are often not classified, although they are of utmost importance in goal setting and decision making in SLP intervention (Light and McNaughton, 2015). The ICF-CY framework can also provide SLPs the opportunity to value the positive aspects of functioning (in addition to the limitations), although this is often not done: 'For different users, it might be appropriate and helpful to add other kinds of information to the coding of each item … At the user's discretion, coding scales can be developed to capture the positive aspects of functioning'(WHO, 2007: 234–235).

In the present study, we used ICF-CY in a case-based approach in SLP as a general conceptual framework to examine the contribution of significant factors (e.g. environmental and personal) on communication performance in six young children with Down syndrome, controlling for mental age and/or the level of their lexical development. A case-based approach involves an in-depth, and detailed examination of a participant, as well as its related contextual conditions. In this multiple case study, the six children are not considered to be a group of children with Down syndrome (for group analyses, see chapter 4), but are analysed as six separate cases. Our research is guided by the following question: Does the ICF-CY provide a suitable framework to characterize the unique communication performance of children with Down syndrome and significant others in their direct social environment?

# Method

## Participants

Parents of children with DS were recruited via social media of the Dutch parent association *Stichting Downsyndroom* and included in the study if their child met the following inclusion criteria: (a) chronological age between 2;0 and 7;6 years, and (b) Dutch-speaking home environment, (c) parents report problems with their child's communication performance. No exclusion criteria were taken into account. Six parents agreed with participation in this multiple- case study. All children were genetically diagnosed with Trisomy-21, the most common form of Down syndrome. The study was approved by an Institutional Review Board (NL38926.091.12).

## Procedures

The data collection focussed on identifying individual and environmental strengths and weaknesses in functioning in several areas related to communication performance, including attention regulation, working memory, cognition, metalizing (socio-emotional) capacity, orientation and adaptive skills, production and comprehension of language, speech or other representational forms of language, and vocabulary development (see Denes, 2011). First, parents were sent multiple questionnaires pertaining to communication performance (see Table 1). Second, the children's significant communicative partners, identified by the Social Networks Inventory (Blackstone & Hunt-Berg, 2003), were interviewed, including parent(s), teacher or day care providers, SLPs, and other relevant professionals. Semi-structured guided interviews examined communication-related activities and participation, as in Power, Anderson and Togher (2011). Thirdly, medical and/or institutional files were analysed to obtain test scores (e.g. mental age, auditory or visual abnormalities) and clinical reports. Fourth, extensive observations, both in structured and unstructured situations, were used to map environmental factors (Schiariti et al., 2014). At home (e.g., mealtime and play situations), at school, during day care and/or therapy sessions (e.g., instants of instruction, interactions with peers, teacher or care, play situations), videotapes were made and analysed with a structured observation instrument (i.e., Communicatieve Intentie Onderzoek, see Table 1 for a brief description). Fifth and finally, (standardized) tests were administered (see Table 1). Dependent on the attention span and motivation of the child, tests were administered in one or multiple sessions. If multiple sessions were needed, these were always within a time frame of a maximum of two weeks. Appendix A shows an overview of the assessment instruments linked to the identified ICF-CY codes, following the

case description provided by McLeod (2006). Data were collected by final year undergraduate SLP students. They received extensive training in communication skills with children with intellectual disabilities, assessment and interviewing from experts in the field. Protocols for assessment and interviewing are available. Due to space restrictions and the protocols' language (Dutch), these were not added to the appendices.

## Data analysis

The first step in data analysis was to identify the level of communication performance for each individual child: the level of communicative intent (CIO), expressive vocabulary skills (N-CDI), developmental age (Vineland Screener) and social participation (Social Networks). Performance on each of these domains are visualised in Figure 1. The next step was to identify underlying factors possibly contributing to the level of communication performance. Based on all aforementioned methods, the level of impairment, barriers and facilitators in functioning and personal and environmental factors were determined using ICF-CY qualifiers to identify strengths and weaknesses within subjects. Child specific strengths within the activity and participation domains (ICF-CY d-codes) were indicated with criterion-referenced qualifiers in which the child's individual functioning is its own criterion. Unique and shared barriers and facilitators were identified and are described in the Results section. This part of the Results section follows the order of the ICF-CY framework: (a) body structures, (b) body functions, (c) activities and participation, and (e) environmental factors. The level of communication performance and all underlying ICF-CY codes make up an Integrative Profile of Communication Performance (IPCP). These profiles can be found in Figure 1 and Appendix A. The IPCPs of the children are compared and visually inspected to search for possible trends or relationships between the several underlying processes of communication performance and mental and/or vocabulary age. No statistical analysis were conducted due to the small sample size.

Table 1. *Overview of assessment methods. Abbreviations are used in Appendix A.*

| Assesment methods |
|---|
| a. Observations of communicative settings at home (f.e. play), at school/day care, during therapy sessions with professionals (Observation). |
| b. Interviews with parents, teachers/day carers, professionals and other significant communication partners and dossier analysis (Report). |
| c. The Child Behaviour Checklist is a parental questionnaire identifying specific behavioural, emotional, and social problems that characterize preschool children (Achenbach & Rescorla, 2000). |
| d. Subscale working memory from the BRIEF (Huizinga & Smidts, 2012). The BRIEF measures the range of executive function in (pre)school-aged children (BRIEF). |
| e. The Auditory Discrimination Test (Verhoeven & Vermeer, 2001) investigates whether a child can identify sound contrasts in minimal pairs (AD). |
| f. The Auditory Memory Test (Gathercole, Pickering, Hall & Peaker, 2001) measures the longest list of items that a person can remember (AMT). |
| g. The Schlichting Test for expressive language II (Schlichting & Lutje Spelberg, 2010b) measures the level of expressive language skills of young children between 2;0 and 7;0 years (f.e., syntax development and storytelling) (STEL). |
| h. The Schlichting Test for receptive language (Schlichting & Lutje Spelberg, 2010a) measures the level of receptive language skills (STRL). |
| i. The Communication Matrix (Rowland & Fried-Oken, 2010) is an assessment instrument designed for individuals of all ages who function at the earliest stages of communication and who use any form of communication (CM). |
| j. The Communicatieve Intentie Onderzoek [Communicative Intent Assessment] (Van der Meulen, Slofstra-Bremer, & Lutje Spelberg, 2013) is a norm-referenced instrument to observe the preverbal and early stages of the verbal development within a play context with a parent and subsequent with the SLP (CIO). The instrument also scores the competences and limitations in communication performance of communication partners during play/conversation with the child. |
| k. The Quality of Life questionnaire (Centrum voor Consultatie en Expertise, 2010) is specifically constructed for people with (severe) disabilities and can be used to systematically identify specific target areas for health care (QoL). |
| l. The Dutch version of the Receptive One-Word Picture Vocabulary Test (ROWPVT; Brownell, 2000b) measures the receptive vocabulary (RVT). |
| m. The Dutch version of the Expressive One-Word Picture Vocabulary Test (EOWPVT; Brownell, 2000a) measures the expressive vocabulary (EVT). |
| n. Dutch version of the MacArthur-Bates Communicative Development Inventories (Zink & Lejaegere, 2002): parental questionnaire for receptive and expressive vocabulary (N-CDI). |
| o. The Computer Articulation Instrument is a norm-referenced standardized speech production test for children in the age range of 2 to 7 years, that contributes to diagnostic differentiation between phonological disorder and motor speech disorder (Van Haaften et al., 2012) (CAI). |
| p. Spontaneous Speech Analysis of samples during spontaneous speech (SSA). |
| q. The Screening instrument of Emergent Literacy skills (Vloedgraven Keuning & Verhoeven, 2009) identifies the level of phonological awareness and receptive and expressive letter knowledge (SEL). |
| r. The Bridge: Emergent literacy (Pierce, Summer & O'DeKirk, 2005) is a portfolio rating scale of oral language and emergent literacy behaviours (Bridge). |
| s. The Social Networks Inventory (Blackstone & Hunt-Berg, 2003) is an instrument to identify existing relationships of an individual and to identify opportunities and barriers to participation in life activities (SN). |
| t. The Vineland Screener 0-6 measures the level of adaptive functioning of children up to the age of 6 or older people with comparable levels of functioning (Scholte et al., 2008). |

## ICF-CY Coding

In constructing our assessment framework, we combined the core sets of ICF(-CY) codes formulated for communication disability by Simeonsson (2003), speech impairment (McLeod, 2006; McLeod & McCormack, 2007) and Augmentative and Alternative Communication goal setting (Rowland et al., 2012). For every ICF-CY code, we indicated applicable methods of assessment, according to the rules proposed by Cieza et al. (2002): we indicated which assessment instruments are relevant to the separate ICF-CY codes (see Appendix A).

Assigning performance qualifiers in developing children is challenging, since functioning varies widely due to factors as age, task and setting (Kronk, Ogonowski, Rice, & Feldman, 2005). Challenges occur in operationalizing the qualifier level, since scale steps are not well defined within the ICF-CY itself (Simeonsson, 2014). Thus, we followed the coding guidelines by Simeonsson, Lee, Granlund, & Bjorck-Akesson (2010, p. 39) in which they determine qualifiers based on standard scores, scaled scores, T-scores, percentile ranks and standard deviation units. In those instances where clinical judgement was involved, we followed the procedure as described by Kronk et al. (2005). To increase interrater reliability, Kronk et al. proposed to collapse the performance qualifiers into two categories: mild-moderate and severe-complete problems.

### *Interrater reliability of ICF-CY coding*

A fundamental prerequisite for use of the ICF-CY with children is the demonstration of reliable coding within a developmental perspective (Kronk et al., 2005, p. 978). The joint probability of agreement between three independent raters on all ICF-CY codes used in the IPCPs was 77%, with a range in agreement between two out of three raters ranging from 80 – 92 %. Intraclass Correlation Co-efficient (absolute agreement) over the three raters was .63, 95% CI [.54 - .71], indicating a substantial agreement between raters (see Viera & Garrett, 2005). The individual case profiles as provided in the results section were then constructed in consensus with these three raters. Finally, these profiles were discussed with the parents. They indicated on a 5-point scale (1 = completely incorrect, 5 = completely correct) whether a) the provided profile was commensurable with their own experiences, and b) all important characteristics of their child's functioning were embedded in the profile. All parents scored a 4 or 5 on both questions, indicating parents could (highly) identify their child with the presented IPCPs.

# Results

## Participants

The six children (fictitious names) had a mean chronological age of 3;9 years (*SD* = 8 months) and boys and girls were equally represented. Mental ages (*M* = 2;3 years, *SD* = 4 months), expressive vocabulary age (*M* = 1;9 years, *SD* = 2 months), and receptive vocabulary age (*M* = 1;7 years, *SD* = 3 months) of each of the children is recorded in Table 2. From this table, we can conclude that the six children with Down syndrome are each comparable to at least one of the other children on either chronological, mental, and/or expressive vocabulary age. To provide more insight into the children involved, the communication performance of the children is described below in more detail.

Table 2. *Descriptives of the Six Participants.*

| Child | Gender[a] | ICD-10 code[b] | Chronological age[c] | Mental age[c] | Expressive vocabulary age[c] | Receptive vocabulary age[c] |
|---|---|---|---|---|---|---|
| Jonas[d] | M | Q90.0 T21 MN | 4;9 | 1;9 | 1;7 | 1;5 |
| Mary[d] | F | Q90.0 T21 MN | 4;4 | 1;11 | 1;6 | 1;5 |
| Toby[d] | M | Q90.0 T21 MN | 3;1 | 2;0 | 1;9 | 1;6 |
| Erna[d] | F | Q90.0 T21 MN | 3;9 | 2;6 | 1;8 | 1;7 |
| Fenna[d] | F | Q90.0 T21 MN | 3;2 | 2;6 | 1;9 | 1;7 |
| Nick[d] | M | Q90.0 T21 MN | 3;5 | 2;7 | 1;11 | 2;0 |

*Note*. a M = male; F = female. b T21 MN = Trisomy 21, meiotic nondisjunction. c Ages are in years [years;months]. d Used names are fictive.

### *Jonas*

Jonas is severely impaired in both his reception and expression of spoken language. He mostly relies on body language, gestures and vocalizations and is able to speak a few words, although often unintelligible. Symbol use (such as graphic symbols) is only very slowly emerging and Jonas does not combine symbols. He uses symbols especially to refuse or reject something or to make choices (level of use is based on the Communication Matrix; Rowland & Fried-Oken, 2010). When investigating his social network (see Blackstone & Hunt-Berg, 2003), we notice that his communication partners are only situated in the circles of direct family and professionals. His communication via body language, gestures and vocalizations is indicated as being often effective by his communication partners, with parents understanding him more often than the professionals. Communication partners mostly use spoken language and (natural) signs, which is often not very effective.

Professionals sometimes also use graphic symbols, however, this is often not effective. Parents indicate that professionals that are involved do not discuss goals and do not synchronize with each other and with parents in the ways they communicate with Jonas.

### *Mary*

Mary is severely impaired in her expression and reception of spoken language and in her expression of gestural language. Her main strength is producing body language. Mary is able to produce some onomatopoetic words. She does not yet combine two symbols into multiple word utterances and makes no use of abstract symbols. Some concrete symbols are emerging. Within direct family circles and with classmates, she mostly communicates via vocalizations, body language and natural signs. With professionals, she predominantly communicates via objects and graphic symbols. Parents as well as professionals indicate they often can understand Mary very well. Communication partners generally use spoken language, signs, and body language. Partners indicate that especially when they combine these communication modes, Mary can understand their messages.

### *Toby*

Toby is impaired in both his reception and expression of spoken language. Toby's speech is emerging, he is able to speak a few words. In general he communicates via manual signs and through vocalisations. Vocalising when aware of another person in the proximal environment is one of his most important strengths, through which Toby is able to initiate social interactions. He does not yet combine symbols into two- or three symbol combinations. When investigating his social network, we can see that his communication partners are only situated in the direct family and circle of professionals, with the exception of only one classmate. Toby does not differentiate in the ways in which he communicates with these different partners. Within his direct family the communication modes are often successful, where this is only sometimes the case with other communication partners. Communication partners mostly use signs and spoken language to communicate with Toby, which he can often understand.

### *Erna*

Erna is severely impaired in her expression of spoken language. She only speaks a few words and is often unintelligible. She does not yet combine two symbols into multiple word utterances and mostly relies on her strengths in gestural and body language. Her abstract symbol use is developing. Remarkable are her skills in

comprehending simple spoken messages. When investigating the social network of Erna, we can see that her communication partners are situated in the direct family and professionals, with the exception of two classmates. Erna does not distinguish in the way she communicates with these different partners, although her communication modes are often not effective with some professionals and friends. Parents indicate they understand her very well. Communication partners mostly use a combination of speech, signs and body language.

### *Fenna*

Although Fenna is severely impaired in her expression of spoken language, her expression of gestural language is a relative strength as is her reception of spoken language. Fenna's speech is emerging, she is able to speak a few words, although communication partners indicate she is often unintelligible. Combinations of symbols into two-symbol combinations are also emerging. When investigating her social network, we can see that her communication partners are situated in all circles, except for unfamiliar partners. She has the largest number of communication partners of the six children. Fenna does not differentiate in the ways in which she communicates with these different partners. No differences in effectiveness of used communication modes amongst different partners are seen, they all indicate her communication as effective. Only her classmates seem to understand her less than other partners do. Communication partners mostly use spoken language, signs and body language.



### *Nick*

Nick is able to speak, although often unintelligible and he slowly emerges to combine symbols into two and three symbol combinations. However, Nick is severely impaired in his mental functions that organize semantic and symbolic meaning, grammatical structure and ideas for the production of messages. Mostly he communicates via body language, facial expressions, vocalizations and spoken language. He especially uses these communication modes to seek help. When investigating his social network, we can see that his communication partners are only situated in the direct family and professionals. Nick has the smallest number of communication partners of the six children. Nick does not differentiate in the way he communicates with these different partners, although his communication modes are only sometimes effective with his parents. Communication with his brother is often difficult, which seems to be related to his brother's diagnosis of autism and ADHD. Communication partners mostly use spoken language, facial expressions and body language, modes that Nick

understands well. All communication partners mention behavioural problems when Nick is not able to express himself properly.

## Communication performance

Figure 1 shows the level of communication performance of the six children. This figure shows that although the measured level of communicative intent is comparable for some children, they differ in expressive vocabulary skills. Where Jonas and Toby show expressive skills somewhat comparable to their mental age, Erna, Fenna, and Nick show expressive skills that lag 5 to 9 months behind their mental age. Data on participation within the circles of Social Networks indicate that Fenna, the child with the highest communicative intent scores, but not with the highest vocabulary or mental age, also showed the highest number of communication partners and successful communication performance in all settings. The level of effective communication performance within the circles can not directly be explained by the expressive vocabulary skills or mental ages, given that Nick scores highest on both measurements, but lowest on the level of participation within social circles. Overall, we may conclude that we can not explain communication performance solely based on the level of communicative intent, mental age and expressive vocabulary scores. This also implicates that the level of communicative intent in combination with the mental age does not solely account for the level of expressive vocabulary skills. For example, Toby and Fenna show comparable expressive vocabulary ages and differ on their level of communicative intent and mental age. To identify underlying factors in communication performance, we will further discuss similarities and differences between the six subjects on identified ICF-CY codes.

## Between subject similarities

Examining the IPCPs, similarities for most of the children were identified. These similarities will be described separately for (a) body structures, (b) body functions, (c) activities and participation, and (e) environmental factors. See Table 3 for an abbreviated version of the IPCPs and Appendix A for the complete IPCPs.

### *Body structures*

Three out of the six children showed no impairments in body structures (s110 – s5408), where the other three children showed a mild impairment in one body structure.

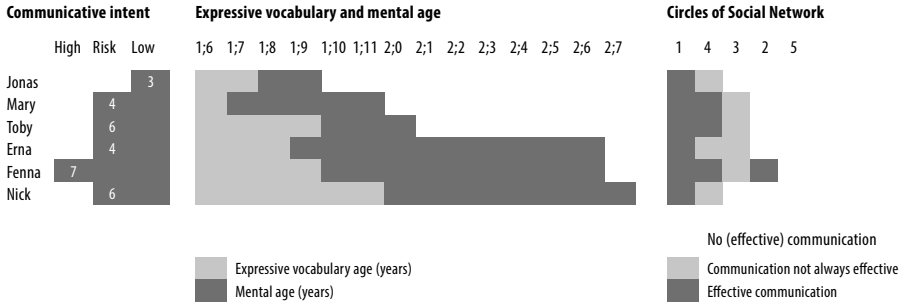



Figure 1. The level of communication performance of the six children.

*Note*. Referential data derived from 'The Communicatieve Intentie Onderzoek (CIO)' indicates whether the level of communicative intent is mental age appropriate (i.e. Wechsler-score of seven or up), at possible risk for developing problems (i.e. Wechsler-score of four, five or six) or very low and problematic (i.e. Wechsler-score of three or below). The expressive vocabulary age is based on the N-CDI (Zink & Lejaegere, 2002), in which both spoken and signed vocabulary is assessed. The participation within the circles of social networks is based on the Social Networks Inventory (Blackstone & Hunt-Berg, 2003) and observations in different settings: (1) family members/life partners, (2) friends, (3) acquaintances, (4) paid professionals, and (5) unfamiliar people.



## *Body functions*

Most children show no impairment in consciousness functions (b110), energy level (b1300), motivation (b1301), speech discrimination (b2304), proprioceptive functions (b260), respiration and ingestion functions (b440, b510). On the other hand, most children show severe impairments in orientation skills (b1140, b1141, b11420, b11421), attention skills (b1400, b1402), short term memory and retrieval and processing of memory (b1440, b1442), organisation and planning (b1641), expression of spoken language (b16710), production of intelligible words and sentences (b3204) and muscle tone functions (b735). Since body functions as attention, memory and planning play a conditional role in communication performance, and given the similarities in impairments across children, it could be expected that all six children score comparable on language and intellectual outcomes. When a direct relation is extant, we may expect that the children with a higher developmental or vocabulary age should show less impairments in body functions. This is not the case, however, even though in some visual aspects we see a trend that children with better communication performance show less impairments in visual perception (b1561), insight (b1644), visual acuity functions (b2100), and quality of vision (b2102).

Table 3. *Short overview of IPCPs.*

| Children | | | | | | | |
|---|---|---|---|---|---|---|---|
| **ICF-CY code** | **J** | **M** | **T** | **E** | **F** | **N** | **Method of assessment (see App. A)** |
| **d3 COMMUNICATION** | | | | | | | |
| **d310-d329 COMMUNICATING - RECEIVING** | | | | | | | |
| d3100 Responding to the human voice | - | 0 | + | + | + | + | Observation |
| d3101 Comprehending simple spoken messages | - | - | - | + | + | + | Observation, CIO, RVT |
| d3102 Comprehending complex spoken messages | -- | -- | -- | -- | - | 0 | Observation, CIO, STRL |
| d3150 Communicating with - receiving - body gestures | 0 | + | + | - | + | - | Observation, SN, report, CIO |
| d3151 Communicating with - receiving - general signs and symbols | - | - | - | - | 0 | 0 | Observation, SN, report, CIO |
| d3152 Communicating with - receiving - drawings and photographs | - | - | - | 0 | 0 | + | Observation, SN, report, STEL, EVT, CIO |
| d320 Communicating with – receiving – formal sign language messages | - | + | 0 | 0 | + | 0 | Observation, SN, report, CIO |
| d325 Communication with - receiving - written messages | -- | -- | -- | -- | -- | -- | Observation, SN, report |
| **d330-d349 COMMUNICATING - PRODUCING** | | | | | | | |
| d330 Speaking | -- | -- | -- | -- | - | - | Observation, report, SN, CM, CIO |
| d331 Pre-talking | -- | 0 | - | 0 | + | + | Observation, report, SN, CM, CIO |
| d332 Singing | ? | -- | ? | -- | + | ? | Observation, report |
| d3350 Producing body language | + | 0 | + | - | + | 0 | Observation, report, SN, CM, CIO |
| d3351 Producing signs and symbols | -- | -- | - | -- | - | - | Observation, report, SN, CM, CIO |
| d3352 Producing drawings and photographs | -- | -- | - | - | - | - | Observation, report, SN, CM |
| d340 Producing messages in formal sign language | - | - | 0 | - | 0 | NA | Observation, report, SN, CM, CIO |
| d345 Writing messages | -- | -- | -- | -- | -- | -- | Report, Bridge |
| **d350-d369 CONVERSATION AND USE OF COMMUNICATION DEVICES AND TECHNIQUES** | | | | | | | |
| d3500 Starting a conversation | -- | -- | -- | -- | -- | - | Observation, report |
| d3501 Sustaining a conversation | -- | -- | -- | -- | -- | -- | Observation, report |
| d3502 Ending a conversation | -- | -- | -- | -- | -- | -- | Observation, report |
| d3503 Conversing with one person | -- | - | - | -- | 0 | + | Observation, report |
| d3504 Conversing with many people | -- | -- | -- | -- | 0 | - | Observation, report |
| d355 Discussion | -- | -- | -- | -- | -- | -- | Observation, report |
| d360 Using communication devices and techniques | NA | NA | NA | NA | NA | NA | |

Note. Body functions and structures: 0 = no problem (.0); - = mild (.1) to moderate (.2) problem; -- = severe (.3) to complete (.4) problem; Activities and Participation: 0 = no problem (.0); - = mild (.1) to moderate (.2) problem; -- = severe (.3) to complete (.4) problem, + = relative strength; Environmental factors: -- = severe to complete barrier, - = mild to moderate barrier, 0 = no barrier/facilitator, + = mild to moderate facilitator, ++ substantial to complete facilitator; ? = not observed; NA = not applicable (.9).

### *Activities and participation*

Most children show severe impairments in acquiring information (d132), combining words into phrases (d1331), acquiring syntax (d1332), learning to read (d140), reading and writing (d166, d170), undertaking multiple tasks (d220), comprehending complex spoken messages (d3102) and conversation skills (d3500, d3501, d3502, d355). Although mental age seems to be closely related to those ICF-CY codes such as learning through symbolic or pretend play (d1313, d1314), acquiring single words or meaningful symbols (d1330), rehearsing (d135), acquiring basic or complex concepts (d1370, d1371), acquiring skills to recognize symbols (d1400), thinking (d163), and pretending (d1630), this trend is not that apparent for the identified levels of communication performance. On the other hand, most children share strengths in copying (d130), responding to the human voice (d3100), maintaining a sitting position (d4153), moving around within the home or other buildings (d4600, d4601), eating (d550), drinking (d560), and initiating social interactions (d71040). Regarding the communication domain (d3) within ICF-CY, we see a trend with mental age concordant with the language outcomes with receptive language (d310-d329), but not with expressive language (d330-d349).



### *Environmental factors*

General products and technology for personal use in daily living and for play (e1150, e11520) are available for all six children. Also health and education services (e580, e585) are provided for all six children, as stated in Dutch law. Where the accessibility to products and health and education services is equivalent for all six children, the differences in communication performance do not pertain to these environmental influences. Immediate family (e310) and health professionals (e355) are indicated as facilitators in five out of six children. The lack of (effective communication with) communication partners in the circles of friends (e320) and peers or acquaintances (e325) is also prominent in five out of six children.

## Between subject differences

In the previous section we primarily portrayed similarities between the six children. In this section we will focus on individual differences.

### *Body functions*

Where visual perception (b1561) is often mentioned as a strength in children with Down syndrome, only Fenna and Nick show no impairment. While both show impairments in tactile perception (b1564) the other four children do not. Auditory

perception (b1560) is impaired in Toby, Erna, and possibly in Jonas, but not in the other three children. Four children show no to mild impairments in adaptation skills (b1250, b1253, b1254, b1255), Jonas (the child with the lowest mental age) and Nick (the child with the highest mental age) show mild to severe problems with adaptability.

### *Activities and participation*

Where all six children show severe impairments in attention skills, three children show better focused attention on human touch, face and voice (d1600), where five show better focused attention to interesting objects (d1608). Where five children can walk or move around within their surroundings (d450 to d4602) easily, Toby lacks the skills for doing so; however this is facilitated by assistive products for mobility (e1201). Two children show mild to severe impairments in all indicated general tasks and demands (d2).

### *Environmental factors*

If we inspect the role of the environmental factors, we see that for Nick most communication partners and especially his immediate family (e310) are a great barrier for his development. Nick has a brother with a diagnosis of Autism Spectrum Disorder. The care for two children with a disability is a heavy burden for the parents, especially because they report to have no social network to rely on and to support them. Parents state to be extremely exhausted and therefore not to be able to provide their sons with the best care. At the same time, Nick is the child with the highest mental age in our study. In Erna and Nick we also see that other professionals (e360) are a barrier in the communication performance. We observed that especially their teachers were not equipped in dealing with children with disabilities.

As can be seen in the IPCP's (Appendix A), all six children share severe impairments in many ICF-CY domains, but show different communication performance skills. Unique factors to a child may play an important role in their communication performance. Table 4 provides an overview of unique barriers and unique facilitators.

Table 4. *Unique barriers and facilitators of the six children.*

| Jonas | Mary | Toby | Erna | Fenna | Nick |
|---|---|---|---|---|---|
| b1250 Adaptability | b1402 Dividing attention | b440 Respiration functions | b230 Hearing functions | b1564 Tactile perception | b125 Dispositions and intra-personal functions |
| b1400 Sustaining attention | b1442 Retrieval and processing of memory | d450-d469 Walking and moving | s250 Structure of the middle ear | b1568 Sensory integration | b130 Energy and drive functions |
| d1330 Acquiring single words or meaningful symbols | b1561 Visual perception | e1201 Assistive products and technology for indoor/outdoor mobility | d2500 Accepting novelty | b235 Vestibular functions | b1400 Sustaining attention |
| d3100 Responding to the human voice | b210 Seeing functions | e315 Extended family | | d177 Making decisions | d571 Looking after one's safety |
| e355 health professionals | b3202 Production of tones | | | d2501 Responding to demands | e310 Immediate family |
| | b7602 Coordination of voluntary movements | | | d2502 Approaching persons or situations | e340 Personal care providers |
| | | | | d571 Looking after one's safety | d1311 Learning through actions by relating two or more objects |
| | | | | e325 Acquaintances, peers | d1401 Acquiring skills to sound out written words |
| | | | | | d3152 Communicating with – receiving – drawings and photographs |
| | | | | | d3503 Conversing with one person |

*Note.* Orange = barriers; Green = facilitators.

## Discussion

Contrary to the proclaimed definition of communication performance as stated by the WHO, the level of communication performance can't simply be explained uniquely by the level of communicative intent, expressive skills, mental age, and social participation. Although our six children shared either a mental and/or expressive vocabulary age and/or level of communicative intent, the communication performance was very divergent. All children showed similar but also unique personal and environmental factors that play a role in their communication performance. It seemed impossible to predict the level of communication performance solely on the basis of the number of strengths and weaknesses in body structures, body functions, activity and participation domains and the environmental factors from the ICF-CY framework.

For example, all children with Down syndrome in the present study experienced weaknesses in several body functions. Most children showed a weakness in sustained attention, concentration, short-term memory, undertaking of multiple tasks, orientation skills and adaptability. Problems found in sustained attention and short-term memory are not surprising, since children with DS are known for their problems in these specific processes (e.g., Brown et al., 2003; Hick, Botting, & Conti-Ramsden, 2005). Provided the interdependency of factors (in terms of ICF-CY domains), it is therefore not surprising that the language and communication development of the children in this study is (severely) distorted, yet to a different extent. As can be seen in the IPCPs, each of the six children with Down syndrome has its own facilitating factors, both personal and within the environment. Importantly to consider when using the ICF-CY, is that the current one-dimensional view of environmental factors as either facilitators or barriers may be a shortcoming of the ICF-CY since their impact may be highly dependent on the context (Adolfsson, 2011). We agree with Adolfsson, in the sense that, for example, only indicating that immediate family has a moderate facilitating role (e310+2) is not informative for intervention planning. Rather, we should determine more closely in which situations, contexts and/or activities the immediate family has a facilitating role. Even more importantly, it should be determined why this is facilitating the child's functioning and its communication performance. In the present study, no environmental or personal factors could be identified as the most important factors contributing to communication performance. The interdependency of factors makes identification of the contribution of a single factor to communication performance impossible. Given the descriptive character of the ICF-CY framework, the ICF-CY itself seems to provide no clues about these interdependencies between domains influencing the level of communication

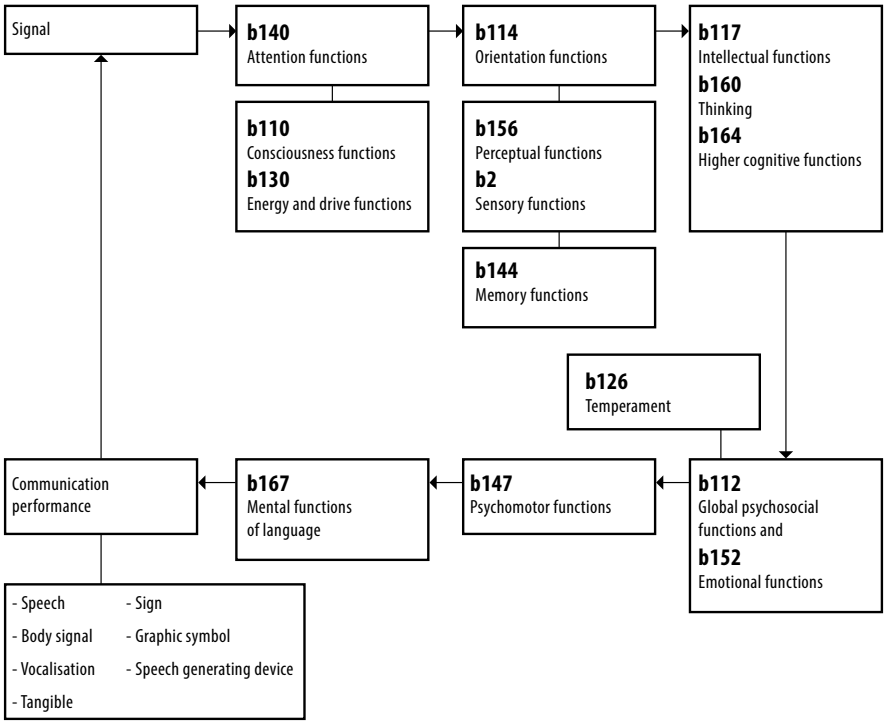



Figure 2. *How communication performance is influenced by ICF-CY body functions.*

performance. The ICF-CY is therefore in its current form not a valuable tool for clinical reasoning in children with Down Syndrome regarding communication performance. We are aware this is not the primary goal of the ICF-CY, and we thus do not deem this as a shortcoming of the ICF-CY. Based on recent neurocognitive studies (see Denes, 2011), theoretical models describing the interdependencies amongst developmental domains in language and communication development (Jackendoff, 2002; Van Balkom, 2009, see Figure 1 in Chapter 1) and our ICF-CY core set for language and communication, we propose an adapted model showing the underlying processes underneath communication performance, which is preceded by mental functions of language which includes a child's lexical development (see Figure 2). This figure provides a reasoning model how these ICF-CY body functions may impact lexical development (b167 Mental functions of language) and communication performance. Environmental factors influencing communication performance can be found in Figure 3.

In the present article we presented a way of profiling communication performance in an integrative profile of communication performance. Our IPCP may provide important input to discuss the (interdependency between) factors influencing communication performance by members of interprofessional teams. Our way of profiling may assist interdisciplinary teams in co-constructing a profile of an individual's functioning and determining intervention goals possibly overlapping multiple professions, adhering to good interprofessional practice (IPP). In agreement with Simeonsson et al. (2006), we conclude that the ultimate goal is to describe the impact of a specific syndrome on various developmental domains for the individual child, and the ICF-CY can be a useful tool in realizing this goal. Assessment frameworks based on the ICF-CY may lead to improved participation and environmental-based goals being set for clients (Dalen et al., 2013; Raghavendra et al., 2007) and their significant communication partners.

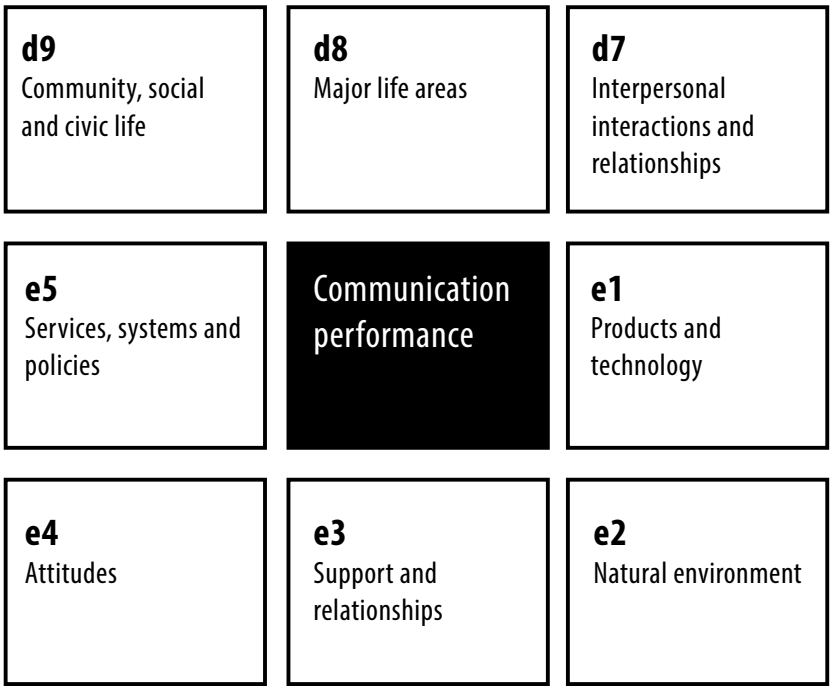



Figure 3. *Environmental influences on communication performance based on ICF-CY.*

We started this article with a description of the Down syndrome behavioural phenotype, a pattern of behavioural strengths and weaknesses specifically associated with Down syndrome. Although this behavioural phenotype describes important factors contributing to communication performance in children with Down syndrome, none of our six children really fit this specific pattern of strengths and weaknesses. Also, this behavioural phenotype does not account for interdependencies between described strengths and weaknesses, and the influence of environmental factors are neglected. For SLPs, it seems warranted that assessment and intervention planning should be based on the reasoning behind our IPCP, rather than on behavioural phenotyping.

## Limitations

Limitations of the present study are: (a) assessment limitations, (b) ICF-CY coding limitations, (c) sample size. The level of communication performance can be determined in several ways: standardized tests, observations and/or parental reports. Standardized tests have the advantage of being based on overt behaviour at a specific time point, and relate to normative data. However, the attention span, which is a problem in the six children with Down syndrome in the present study, is an important factor influencing the assessment (Feldman et al., 2005). Also, these tests have required the child to interact with an unfamiliar adult, possibly influencing the child's responses (Pan et al., 2004), which has been found in children with Down syndrome (Miller et al., 1995). Observations have the advantage of providing insight into the child's actual performance in interaction with communication partners, and are thus potentially more ecologically valid (Pan et al., 2004). However, observations of a conversation with a researcher or a parent may be highly influenced by personality and contextual factors, such as setting and materials used (Feldman et al., 2005; Yont, Snow & Vernon-Feagans, 2003). Children with Down syndrome especially seem to be highly sensitive to these factors. Different communication partners may evoke different communication styles (Pepper & Weitzman, 2004). These factors do not influence data from parental reports. However, there are some studies that have questioned the accuracy of parental reports, particularly in atypical groups, in which parents tend to over- or underestimate the performance of their child (Roberts et al., 1998). In the present study, we tried to overcome these assessment problems. Therefore, we used the method of triangulation and made use of standardized tests, parental reports and observations to determine the functioning of the child.

It is a challenge to assign ICF-CY performance qualifiers in developing children, since functioning varies widely due to factors as age, task, and setting

(Kronk et al., 2005). The ICF-CY itself does not provide the user with well-defined scale steps. Therefore, we followed the coding guidelines by Simeonsson et al. (2010) and Kronk et al. (2005). Although the availability of clear guidelines, assigning qualifiers was still a challenge, especially because of aforementioned assessment limitations, due to which assigning on the basis of standard deviations or percentile scores was not always possible. Several qualifiers thus had to be determined based on clinical reasoning. The present study showed a substantial agreement between three individual raters, with some variation between rater-dyads. All differences were discussed and profiles were adapted in consensus.

A final limitation may be the sample size. Only six children with Down syndrome were studied; as a consequence, no group data can be reported from which conclusions can be drawn (for group data on predictors, see chapter 4). This may limit generalisation of the results of the present study. However, the IPCPs of the six children show substantial differences between children in underlying processes, although their level of communication performance may be comparable. This points out the value of multiple case studies, in which underlying processes can be studied more closely and comprehensively.

## Conclusion

To conclude, although children with Down syndrome can have divergent IPCPs based on ICF-CY codes, the level of communication performance can still be similar. On the other hand, children who share many barriers and facilitators may still develop to varied levels of communication performance. From neurocognitive studies it can be learned learned that limitations in developmental domains as attention and memory are associated with problems in communication performance of children with Down syndrome. The present study shows the myriad of all these influencing factors, internal and external, which make up a profile of strengths and limitations underlying a child's communication performance. Barriers and facilitators, as do child specific strengths and weaknesses, are involved in a reciprocally influencing relationship and are thus interdependent, as shown in the proposed models. When addressing communication performance, both in research and in clinical practice, all these several developmental domains should be incorporated.

# Appendix A

Table A. *Overview of ICF-CY profiles of communication performance of six children with Down syndrome.*

| | Children | | | | | | |
|---|---|---|---|---|---|---|---|
| **ICF-CY code** | **J** | **M** | **T** | **E** | **F** | **N** | **Method of assessment (see App. A)** |
| **B BODY FUNCTIONS** | | | | | | | |
| **b1 MENTAL FUNCTIONS** | | | | | | | |
| **b110-b139 GLOBAL MENTAL FUNCTIONS** | | | | | | | |
| b110 Consciousness functions | 0 | 0 | 0 | 0 | 0 | 0 | Observation |
| b1140 Orientation to time | -- | -- | -- | -- | -- | - | Observation |
| b1141 Orientation to place | -- | -- | - | -- | -- | - | Observation |
| b11420 Orientation to self | -- | -- | -- | -- | 0 | -- | Observation |
| b11421 Orientation to others | -- | -- | -- | -- | - | -- | Observation |
| b117 Intellectual functions | -- | -- | -- | -- | - | - | Report, observation |
| b1250 Adaptability | -- | - | - | 0 | - | -- | Observation, CBCL |
| b1253 Predictability | 0 | - | - | 0 | 0 | -- | Observation, report, CBCL |
| b1254 Persistence | - | - | - | - | - | -- | Observation, report |
| b1255 Approachability | - | 0 | 0 | 0 | 0 | - | Observation, CBCL |
| b126 Temperament and personality functions | - | 0 | 0 | 0 | 0 | - | Observation, CBCL |
| b1264 Openness to experience | - | 0 | 0 | 0 | 0 | - | Observation, CBCL |
| b1300 Energy level | - | 0 | 0 | 0 | 0 | 0 | Observation, CBCL |
| b1301 Motivation | 0 | 0 | 0 | 0 | 0 | - | Observation, report, CBCL |
| b1304 Impulse control | - | - | - | - | - | -- | Observation, report, CBCL |
| **b140-b189 SPECIFIC MENTAL FUNCTIONS** | | | | | | | |
| b1400 Sustaining attention | -- | - | - | - | 0 | -- | Observation, report, CBCL |
| b1402 Dividing attention | - | -- | - | - | - | - | Observation, report, CBCL |
| b1440 Short-term memory | -- | -- | -- | -- | -- | -- | Observation, report, BRIEF, AMT |
| b1442 Retrieval and processing of memory | - | -- | - | - | - | - | Observation, report, BRIEF, STEL |
| b1560 Auditory perception | ? | 0 | - | - | 0 | 0 | Report, observation, QoL |
| b1561 Visual perception | - | -- | - | - | 0 | 0 | Report, observation, QoL |
| b1564 Tactile perception | 0 | 0 | 0 | 0 | -- | - | Report, observation, QoL |

| ICF-CY code | J | M | T | E | F | N | Method of assessment (see App. A) |
|---|---|---|---|---|---|---|---|
| b1568 Perceptual functions, otherwise specified: sensory integration | -- | 0 | 0 | 0 | -- | - | Report, observation |
| b1600 Pace of thought | -- | -- | -- | - | - | - | Not observed |
| b163 Basic cognitive functions | -- | -- | -- | -- | - | - | Report |
| b1640 Abstraction | -- | -- | - | - | - | - | Observation, CM |
| b1641 Organization and planning | -- | -- | -- | -- | -- | -- | Observation, report |
| b1644 Insight | -- | -- | - | - | - | 0 | Observation, report, CIO |
| b1646 Problem solving | -- | -- | - | - | - | - | Observation, report, CIO |
| b16700 Reception of spoken language | -- | -- | -- | - | - | - | Observation, Report, RVT, LEX, CIO, SN |
| b16702 Reception of sign language | - | - | 0 | 0 | 0 | 0 | Observation, report, RVT, LEX, CIO, SN |
| b16710 Expression of spoken language | -- | -- | -- | -- | -- | - | Observation, report, EVT, N-CDI, CIO, CM, SN |
| b16712 Expression of sign language | - | - | 0 | 0 | 0 | 0 | Observation, report, EVT, N-CDI, CIO, CM, SN |
| b16713 Expression of gestural language | - | - | 0 | 0 | 0 | 0 | Observation, report, CIO, CM, SN |
| b1672 Integrative language functions | ? | ? | ? | ? | ? | ? | Not observed |
| b176 Mental function of sequencing complex movements | -- | -- | - | - | - | -- | Observation, report |
| b180 Experience of self and time functions | ? | ? | ? | ? | ? | ? | Not observed |
| **b2 SENSORY FUNCTIONS AND PAIN** | | | | | | | |
| **b210–b229 SEEING AND RELATED FUNCTIONS** | | | | | | | |
| b2100 Visual acuity functions | -- | -- | -- | -- | 0 | 0 | Report |
| b2101 Visual field functions | 0 | - | 0 | 0 | 0 | 0 | Report |
| b2102 Quality of vision | - | -- | - | 0 | 0 | 0 | Report |
| b2152 Functions of external muscles of the eye | 0 | - | 0 | 0 | 0 | 0 | Report |
| **b230-b249 HEARING AND VESTIBULAR FUNCTIONS** | | | | | | | |
| b230 Hearing functions | - | -- | - | -- | -- | 0 | Report, AD |
| b2300 Sound detection | - | 0 | - | -- | 0 | 0 | Report, observation |
| b2301 Sound discrimination | -- | -- | - | -- | -- | 0 | Report, AD |
| b2302 Localisation of sound source | 0 | 0 | 0 | -- | 0 | 0 | Report, observation |
| b2304 Speech discrimination | 0 | 0 | 0 | 0 | 0 | 0 | Report, observation |
| b235 Vestibular functions | 0 | 0 | 0 | 0 | - | 0 | Report, observation |
| **b250-b279 ADDITIONAL SENSORY FUNCTIONS** | | | | | | | |
| b260 Proprioceptive function | 0 | 0 | 0 | 0 | 0 | 0 | Observation, report |

| | | | | | | | |
|---|---|---|---|---|---|---|---|
| **b3 VOICE FUNCTIONS** | | | | | | | |
| b310 Voice functions | 0 | - | 0 | - | - | 0 | Observation, report, CAI, SSA |
| b3200 Production of vowels | 0 | - | - | - | 0 | 0 | Observation, report, CAI, SSA |
| b3201 Production of consonants | - | -- | - | -- | - | - | Observation, report, CAI, SSA |
| b3202 Production of tones | 0 | -- | 0 | - | 0 | 0 | Observation, report, CAI, SSA |
| b3203 Production of syllable shapes | -- | -- | -- | -- | - | - | Unknown |
| b3204 Production of intelligible words and sentences | -- | -- | -- | -- | - | -- | Observation, report, CAI, EVT, SSA |
| b330 Fluency and rhythm of speech functions | NA | NA | NA | NA | -- | -- | Observation, report, SSA |
| b3401 Making a range of sounds | 0 | - | 0 | - | 0 | 0 | |
| b4 FUNCTIONS OF THE CARDIOVASCULAR, HAEMATOLOGICAL, IMMUNOLOGICAL AND RESPIRATORY SYSTEMS | | | | | | | |
| b440 Respiration functions | 0 | 0 | - | 0 | 0 | 0 | Report |
| b5 FUNCTIONS OF THE DIGESTIVE, METABOLIC AND ENDOCRINE SYSTEMS | | | | | | | |
| b510 Ingestion functions | 0 | 0 | 0 | 0 | 0 | 0 | Observation, report |
| b7 NEUROMUSCULOSKELETAL AND MOVEMENT-RELATED FUNCTIONS | | | | | | | |
| b710 Mobility of joint functions | 0 | 0 | 0 | -- | -- | 0 | Observation, report |
| b735 Muscle tone functions | - | - | -- | -- | -- | - | Observation, report |
| b7602 Coordination of voluntary movements | 0 | - | 0 | 0 | 0 | 0 | Observation, report |
| S BODY STRUCTURES | | | | | | | |
| s110 Structure of brain | ? | ? | ? | ? | ? | ? | Not observed |
| s220 Structure of eyeball | 0 | 0 | 0 | 0 | 0 | 0 | Observation |
| s240 Structure of external ear | 0 | - | 0 | 0 | 0 | 0 | Observation |
| s250 Structure of middle ear | 0 | 0 | 0 | - | 0 | 0 | Report |
| s260 Structure of inner ear | ? | ? | ? | ? | ? | ? | Not observed |
| s5408 Structure of intestine, other specified | - | 0 | 0 | 0 | 0 | 0 | Report |
| D ACTIVITIES AND PARTICIPATION | | | | | | | |
| **d1 LEARNING AND APPLYING KNOWLEDGE** | | | | | | | |
| **d110-d129 PURPOSEFUL SENSORY EXPERIENCES** | | | | | | | |
| d110 Watching | - | - | 0 | - | + | 0 | Observation, report, CIO, EVT |
| d115 Listening | - | + | + | - | + | + | Observation, report, EVT, CIO |
| d1201 Touching | -- | -- | 0 | - | 0 | 0 | Observation, report |

| ICF-CY code | J | M | T | E | F | N | Method of assessment (see App. A) |
|---|---|---|---|---|---|---|---|
| **d130-d159 BASIC LEARNING AND ACQUIRING KNOWLEDGE** | | | | | | | |
| d130 Copying | 0 | 0 | + | + | + | + | Observation, CIO, CAI |
| d1310 Learning through simple actions with a single object | - | + | + | - | + | + | Observation |
| d1311 Learning through actions by relating two or more objects | -- | - | 0 | -- | 0 | + | Observation |
| d1313 Learning through symbolic play | -- | -- | + | ? | + | + | Observation |
| d1314 Learning through pretend play | -- | -- | - | 0 | + | + | Observation |
| d132 Acquiring information | -- | -- | -- | -- | - | - | Observation, CM, N-CDI |
| d1330 Acquiring single words or meaningful symbols | -- | 0 | 0 | + | + | + | N-CDI, EVT, RVT, CM |
| d1331 Combining words into phrases | -- | -- | -- | -- | - | - | N-CDI, CM, Observation |
| d1332 Acquiring syntax | -- | -- | -- | -- | -- | - | N-CDI, Observation, STEL |
| d135 Rehearsing | -- | -- | -- | - | + | + | Observation, Report |
| d1370 Acquiring basic concepts | -- | -- | - | + | + | + | N-CDI, EVT, RVT |
| d1371 Acquiring complex concepts | -- | -- | -- | - | - | 0 | N-CDI, EVT, RVT |
| d140 Learning to read | -- | -- | -- | -- | -- | - | Bridge, SEL |
| d1400 Acquiring skills to recognize symbols | -- | - | - | + | + | + | Bridge, SEL |
| d1401 Acquiring skills to sound out written words | -- | -- | -- | -- | -- | + | Bridge, Report, Observation |
| d1402 Acquiring skills to understand written words and phrases | -- | -- | -- | -- | -- | - | Bridge, Report, Observation |
| d1550 Acquiring basic skills | + | + | + | - | + | + | Observation, report |
| d1551 Acquiring complex skills | -- | -- | - | -- | 0 | - | Observation, report |
| **d160-d179 APPLYING KNOWLEDGE** | | | | | | | |
| d160 Focusing attention | -- | 0 | 0 | -- | 0 | - | Observation, report, CBCL |
| d1600 Focusing attention on human touch, face and voice | - | + | - | - | + | + | Observation |
| d1601 Focusing attention to changes in the environment | -- | 0 | -- | -- | - | - | Observation |
| d1608 Focusing attention, otherwise specified: to interesting objects | + | + | + | + | + | - | Observation |
| d163 Thinking | -- | -- | -- | -- | 0 | 0 | Observation |
| d1630 Pretending | -- | -- | - | -- | + | + | Observation |
| d166 Reading | -- | -- | -- | -- | -- | - | Bridge, SEL, report |
| d170 Writing | -- | -- | -- | -- | -- | -- | Bridge, report |
| d175 Solving problems | -- | -- | 0 | -- | + | + | Observation, CIO |
| d177 Making decisions | -- | -- | 0 | -- | + | - | Observation, report, CM |

| | | | | | | | |
|---|---|---|---|---|---|---|---|
| **d2 GENERAL TASKS AND DEMANDS** | | | | | | | |
| d210 Undertaking a single task | - | - | + | 0 | + | + | Observation |
| d220 Undertaking multiple tasks | -- | -- | - | -- | - | - | Observation |
| d230 Carrying out daily routine | - | -- | - | - | 0 | -- | Observation, report, CBCL |
| d2500 Accepting novelty | -- | - | 0 | + | - | -- | Report, CBCL |
| d2501 Responding to demands | -- | - | - | - | + | - | Observation |
| d2502 Approaching persons or situations | - | - | 0 | - | + | - | Observation |
| **d3 COMMUNICATION** | | | | | | | |
| **d310-d329 COMMUNICATING – RECEIVING** | | | | | | | |
| d3100 Responding to the human voice | - | 0 | + | + | + | + | Observation |
| d3101 Comprehending simple spoken messages | - | - | - | + | + | + | Observation, CIO, RVT |
| d3102 Comprehending complex spoken messages | -- | -- | -- | -- | - | 0 | Observation, CIO, STRL |
| d3150 Communicating with - receiving - body gestures | 0 | + | + | - | + | - | Observation, SN, report, CIO |
| d3151 Communicating with - receiving - general signs and symbols | - | - | - | - | 0 | 0 | Observation, SN, report, CIO |
| d3152 Communicating with - receiving - drawings and photographs | - | - | - | 0 | 0 | + | Observation, SN, report, STEL, EVT, CIO |
| d320 Communicating with – receiving – formal sign language messages | - | + | 0 | 0 | + | 0 | Observation, SN, report, CIO |
| d325 Communication with - receiving - written messages | -- | -- | -- | -- | -- | -- | Observation, SN, report |
| **d330-d349 COMMUNICATING - PRODUCING** | | | | | | | |
| d330 Speaking | -- | -- | -- | -- | - | - | Observation, report, SN, CM, CIO |
| d331 Pre-talking | -- | 0 | - | 0 | + | + | Observation, report, SN, CM, CIO |
| d332 Singing | ? | -- | ? | -- | + | ? | Observation, report |
| d3350 Producing body language | + | 0 | + | - | + | 0 | Observation, report, SN, CM, CIO |
| d3351 Producing signs and symbols | -- | -- | - | -- | - | - | Observation, report, SN, CM, CIO |
| d3352 Producing drawings and photographs | -- | -- | - | - | - | - | Observation, report, SN, CM |
| d340 Producing messages in formal sign language | - | - | 0 | - | 0 | NA | Observation, report, SN, CM, CIO |
| d345 Writing messages | -- | -- | -- | -- | -- | -- | Report, Bridge |

| ICF-CY code | J | M | T | E | F | N | Method of assessment (see App. A) |
|---|---|---|---|---|---|---|---|
| **d350-d369 CONVERSATION AND USE OF COMMUNICATION DEVICES AND TECHNIQUES** | | | | | | | |
| d3500 Starting a conversation | -- | -- | -- | -- | -- | - | Observation, report |
| d3501 Sustaining a conversation | -- | -- | -- | -- | -- | -- | Observation, report |
| d3502 Ending a conversation | -- | -- | -- | -- | -- | -- | Observation, report |
| d3503 Conversing with one person | -- | - | - | -- | 0 | + | Observation, report |
| d3504 Conversing with many people | -- | -- | -- | -- | 0 | - | Observation, report |
| d355 Discussion | -- | -- | -- | -- | -- | -- | Observation, report |
| d360 Using communication devices and techniques | NA | NA | NA | NA | NA | NA | Observation, report, CM, SN |
| **d4 MOBILITY** | | | | | | | |
| d4153 Maintaining a sitting position | + | + | + | + | + | - | Observation, report |
| d450 Walking | 0 | + | -- | + | + | + | Observation, report |
| d4550 Crawling | NA | NA | + | NA | NA | NA | Observation, report |
| d4600 Moving around within the home | + | + | - | + | + | + | Observation, report |
| d4601 Moving around within buildings other than home | + | + | - | + | + | + | Observation, report |
| d4602 Moving around outside the home and other buildings | + | + | -- | + | + | + | Observation, report |
| **d5 SELF-CARE** | | | | | | | |
| d550 Eating | 0 | 0 | 0 | 0 | 0 | 0 | Observation, report, CBCL, QoL |
| d560 Drinking | 0 | 0 | 0 | 0 | + | 0 | Observation, report, CBCL, QoL |
| d571 Looking after one's safety | - | - | - | - | + | -- | Observation, report, CBCL |
| **d7 INTERPERSONAL INTERACTIONS AND RELATIONSHIPS** | | | | | | | |
| **d710-d729 GENERAL INTERPERSONAL INTERACTIONS** | | | | | | | |
| d710 Basic interpersonal interactions | -- | - | 0 | - | + | + | Observation, report |
| d71040 Initiating social interactions | - | + | + | 0 | + | + | Observation, report |
| d7107 Taking turns in social interactions | -- | -- | 0 | 0 | - | 0 | Observation, report |
| **d730-d779 PARTICULAR INTERPERSONAL RELATIONSHIPS** | | | | | | | |
| d730 Relating with strangers | - | 0 | + | - | + | 0 | Observation, report, SN |
| d7504 Informal relationships with peers | -- | -- | -- | -- | 0 | -- | Observation, report, SN |
| d7601 Child-parent relationships | - | 0 | + | + | + | + | Observation, report, SN |
| d7602 Sibling relationships | NA | 0 | 0 | NA | - | - | Observation, report, SN |
| d7603 Extended family relationships | ? | ? | 0 | ? | ? | ? | Observation, report, SN |
| **d8 MAJOR LIFE AREAS** | | | | | | | |

| | | | | | | | |
|---|---|---|---|---|---|---|---|
| **d810-d839 EDUCATION** | | | | | | | |
| d815 Preschool education | NA | NA | -- | -- | -- | -- | Observation, report |
| d820 School education | -- | -- | NA | NA | NA | NA | Observation, report |
| **E ENVIRONMENTAL FACTORS** | | | | | | | |
| **e1 PRODUCTS AND TECHNOLOGY** | | | | | | | |
| e1150 General products and technology for personal use in daily living | ++ | ++ | ++ | ++ | ++ | ++ | Observation, report, QOL |
| e11520 General products and technology for play | ++ | ++ | ++ | ++ | ++ | ++ | Observation, report, QOL |
| e1201 Assistive products and technology for indoor/outdoor mobility | NA | NA | + | NA | NA | NA | |
| e1251 Assistive products and technology for communication | -- | + | + | + | NA | NA | Observation, report, QOL |
| **e2 NATURAL ENVIRONMENT AND HUMAN CHANGES TO ENVIRONMENT** | | | | | | | |
| e250 Sound | 0 | 0 | 0 | 0 | 0 | 0 | Observation, report |
| **e3 SUPPORT AND RELATIONSHIPS** | | | | | | | |
| e310 Immediate family | ++ | ++ | + | + | ++ | -- | Observation, report, SN, QOL |
| e315 Extended family | ? | ? | + | ? | ? | -- | Report, SN |
| e320 Friends | -- | -- | -- | -- | - | -- | Report, SN |
| e325 Acquaintances, peers, colleagues, neighbours & community members | -- | -- | - | -- | + | -- | Observation, report, SN |
| e340 Personal care providers and personal assistants | ++ | - | + | 0 | 0 | -- | Report, SN, QOL |
| e350 Domesticated animals | NA | NA | NA | NA | NA | NA | Observation, report |
| e355 Health professionals | - | + | ++ | ++ | ++ | ++ | Observation, report, SN, QoL |
| e360 Other professionals | + | + | + | - | + | - | Observation, report, SN, QoL |
| **e5 SERVICES, SYSTEMS AND POLICIES** | | | | | | | |
| e580 Health services, systems and policies | ++ | ++ | ++ | ++ | ++ | ++ | Report |
| e585 Education and training services, systems and policies | ++ | ++ | ++ | ++ | ++ | ++ | Report |

Note. Body functions and structures: 0 = no problem (.0); - = mild (.1) to moderate (.2) problem; -- = severe (.3) to complete (.4) problem; Activities and Participation: 0 = no problem (.0); - = mild (.1) to moderate (.2) problem; -- = severe (.3) to complete (.4) problem, + = relative strength; Environmental factors: -- = severe to complete barrier, - = mild to moderate barrier, 0 = no barrier/facilitator, + = mild to moderate facilitator, ++ substantial to complete facilitator; ? = not observed; NA = not applicable (.9).

# Chapter 6

## General discussion

# General discussion

This thesis focused on the lexical development of both receptive and expressive vocabulary of young children with Down syndrome. The two main themes of this thesis were the assessment of lexical development in Down syndrome, and identifying its predictors. First of all, the validity of the Dutch version of the MacArthur-Bates Children Developmental Index, a parental questionnaire for measuring lexical size both in spoken and signed modalities, was determined (see Chapter 2). To gain further insight in functional word use in communicative settings, the spontaneous language production of these children was analysed (Chapter 3). In addition, predictors of both receptive and expressive vocabulary development in young children with Down syndrome were determined at the group level (Chapter 4). Finally, strengths and weaknesses in these language and communication-related developmental domains were shown on the individual level within a multiple case-study design, and it was determined how to assess and report on these (Chapter 5).

## Assessment of lexical development

The lexical development of children with Down syndrome, both in research and in clinical practice, is measured in various ways: By means of standardized tests, spontaneous language analyses via observations and/or parental reports. In standardized tests, language is provoked in an isolated situation and may not represent active use of these words for a specific child during the day. Where children with Down syndrome often demonstrate relative strengths in social skills, these skills are used to escape tasks (Daunhauer & Fidler, 2011). These children demonstrate a tendency to be 'cognitively avoidant' and exhibit less willingsness to engage in tasks such as standardized test than children with typical development (Wishart, 1993). Spontaneous language analysis has the advantage to provide insight into how the child actually uses language in interaction with communicative partners, and is thus potentially more ecologically valid than the information derived from the administration of standardized tests (Pan et al., 2004). Parents are another important source to gain insight from, since they have a good understanding of their child's vocabulary use, given the fact that they observe and interact with their child in several contexts on a daily basis (Feldman et al., 2005; Pan et al., 2004). And in the case of children with Down syndrome, parents usually understand imprecise articulated speech or signs of their child better than anyone else. Therefore, the measures used in the present thesis were mostly based on parental reports and observations.

Within these assessments, the multimodal communication skills of children with Down syndrome have to be taken into account. Children with Down syndrome often show impairments in the phonological loop of working memory, which feed into the planning and articulation (i.e., speech) of meaningful words (Jarrold & Baddeley, 2001). To compensate for the difficulties children with Down syndrome encounter in speech production (Caselli et al., 1998), they often switch to the manual modality in early vocabulary development. Manual signing can thus be regarded a more accessible communicative tool for young children with Down syndrome (Özçaliskan, Adamson, Dimitrova, Bailey, & Schmuck, 2016). The early words uttered via manual signs are frequently the first words children with Down syndrome produce by speech (Kouri, 1989), suggesting a tight positive relation between manual sign use and later spoken vocabulary (Özçaliskan et al., 2016). Chapter 2 described how to incorporate the production of signs into the assessment of lexical development in children with Down syndrome via the parental questionnaire N-CDI. This study showed that the concurrent and predictive validity of this adapted version of the N-CDI was good, indicating that parents are an important source for determining the vocabulary size of children with Down syndrome. Özçaliskan et al. (2016) showed that when children's expressive vocabulary size was estimated using both speech and manual signs, the size of the vocabulary of children with Down syndrome is comparable to typically developing children matched on non-verbal mental age. This stands in contrast with the mentioned expressive language problems in the described behavioural phenotype of children with Down syndrome.

When children are reported as knowing words in both the spoken and signed modality, it remains unclear from the adapted version of the N-CDI how they actually use both modalities. A manual sign and a spoken word may be used simultaneously, or manual signs may be used only in situations in which the child's speech is not understood by the communication partner (Vandereet et al., 2011). Also, parental reports offer a parent's extensive knowledge of the entire repertoire of words produced by their children in a wide range of spontaneous contexts, but "are not suitable for capturing lexical organization or access, attention, accuracy in pronunciation, or level of decontextualization in understanding and producing words" (Bello et al., 2014, p. 771). Therefore, data from spontaneous language samples provide important additional input about functional word use, next to the determination of vocabulary size using the adapted N-CDI.

An interesting quantitative issue regarding the production of language is which words children use most often in spontaneous language (Tomasello, 2003). The N-CDI can not provide insights in how and how frequently words are used by children with Down syndrome in daily communicative settings. In Chapter 3, this

information was obtained by observing the spontaneous language use of children with Down syndrome in multiple contexts and with multiple communication partners. The most frequently used words, core vocabulary, throughout several modes of communication can maximize the potential for spontaneous language generation, and are thus an important part of measuring the lexical development of children with Down syndrome. Chapter 3 showed that the types of words in the core vocabulary of young children with Down syndrome appear to be similar in syntactic, semantic, and pragmatic functions to those core words identified by previous researchers of other (a)typically developing populations, although the contribution of content words to the core vocabulary of the children with Down syndrome seems higher than in other populations.

A deficit in the use of function words as compared to content words could be related to the lower production of multi-word utterances (Zampini & D'Odorico, 2011). Children with Down syndrome often omit grammatical markers and verbs more frequently than typically developing peers with comparable mean length of utterances (Hesketh & Chapman, 1998). The modality of communication, the use of manual signs, also influences the findings of relatively more content words than function words in their core vocabulary. In speech-language therapy or Augmentative and Alternative Communication (AAC) interventions, clinicians typically select nouns as the words to teach (Adamson et al., 1992), because these are considered to be easier to teach and assess than function words and are deemed by SLPs of considerable functional use to a communicator (Banajee et al., 2003). In Sign Supported Dutch, manual signs are learned in the immediate everyday context of repeated one-to-one communication with a communication partner. Manual signs are most often used to support speech about an object or action. This creates a highly scaffolded interactive routing centered around referent-symbol mappings (Özçaliskan et al., 2016). Children with Down syndrome may therefore particularly rely on manual signs to convey information about objects and actions in their immediate environment (Dimitrova, Özçaliskan, & Adamson, 2016), explaining the number of signed nouns and the relatively higher number of content over function words. Rehabilitation programs should be focused on the production of function words, since these words are essential for the production of morphologically and syntactically complete utterances (Zampini & D'Odorico, 2011). The next paragraph will focus on the second theme: Predictors of lexical development in children with Down syndrome.

## Predictors of lexical development

Predictors of lexical development in children with Down syndrome were studied at the group level (Chapter 4) and at the individual, case level (Chapter 5). At the group level, the lexical development of expressive vocabulary in children with Down syndrome was predicted by both internal and external factors: (a) the children's adaptive level of functioning predicting both receptive and spoken expressive vocabulary, (b) the children's receptive vocabulary predicting total expressive vocabulary, (c) the maternal educational level predicting early total expressive vocabulary, (d) the children's early levels of communicative intent predicting later spoken expressive vocabulary, (e) the children's phonological and phonemic awareness predicting later spoken expressive vocabulary, and (f) the children's growth in attention skills predicting growth in spoken expressive vocabulary. The receptive vocabulary development could mainly be explained by early levels of receptive vocabulary and their developmental ages. These results closely relate to findings that receptive vocabulary does not show the same weakness as other aspects of language development in children with Down syndrome (Naess et al., 2011) and is more (developmentally) age appropriate.

The study described in Chapter 4 is one of the first longitudinal attempts to monitor the lexical development of children with Down syndrome and is the first to include a wide range of internal and external predictors. The results show that many of the same predictors that play a role in lexical development in typically developing children are also found in our longitudinal study. This adds value to the more recent research base that shows that the language development of children with Down syndrome, when it is measured in both spoken and signed modalities, is delayed rather than deviant from typical development (Polisenka, & Kapalková, 2014). Combined with the results of recent research, this thesis (Chapters 3 and 4) provides evidence that the lexical development of children with Down syndrome thus seems to closely resemble that of typically developing children. Children with Down syndrome only differ in the use of multimodal communication, such as the use of manual signs next to speech. Their production of manual signs predicted the spoken vocabulary size of children one year later (Özçaliskan et al., 2016). In the present thesis, the number of manual signs alone did not predict subsequent spoken vocabulary size. This difference may be due to the age range of the children in the two studies. The children in Özçaliskan's study were on average 2;6 years old, and were thus younger than the children in the present study. The children in the studies of the present thesis, who were between 2 and 8 years-old, may already be beyond the stage in lexical development to see the direct, compensatory effect of manual sign use on later vocabulary development, since most of the

children in the present thesis already started to speak a significant amount of words.

Next to modalities of communication, another important predictor of lexical development of children with Down syndrome lies in their direct environment. In Chapter 4, educational level of mothers was found to be a significant predictor of their children's vocabulary size. This relationship has earlier been found in typically developing children (Taylor et al., 2013) and children with language impairments (Beitchman et al., 2008). Maternal education was also identified by Couzens, Haynes and Cuskelly (2012) to contribute to the cognitive development in individuals with Down syndrome. Direct parental input and parental use of lexical words, which may be influenced by educational levels, was not investigated in the present thesis and is regarded as a limitation of the study in Chapter 4. Kay-Raining Bird and Cleave (2015) found that mothers, irrespective of whether their child had Down syndrome, language impairments or typical development, fine-tune their input in ways that reflect their children's vocabulary knowledge equally well. Mothers of children with Down syndrome were found to talk more than mothers in the other two groups, which may be due to a higher degree of repetition in speech. However, further analyses (MacDonald, New, Cleave, & Kay-Raining Bird, n.d., in Kay-Raining Bird & Cleave, 2015) demonstrated that mothers go beyond simply repeating the same information by providing additional contextual and semantic information. The role of the environment was further specified in Chapter 5. Next to the involvement in interpersonal interactions and relationships that influence receptive and expressive vocabulary development, other environmental factors should be accounted for, such as the use the availability of products and (assistive) technology in the child's environment, acceptance of and attitudes about disabilities and AAC in the community and the availability of services, systems and policies for (health) care and education for children with Down syndrome.

Most predictors of lexical development have been determined within group studies. Group studies, as well as the described Down syndrome behavioural phenotype, can help professionals and families anticipate to areas of relative strength and weakness in children with Down syndrome and plan interventions accordingly without losing sight of individual differences. But, behavioural phenotypes are probabilistic and not deterministic; they can offer guidance about the probability of certain characteristics being present, but in planning interventions for children with Down syndrome, individual differences should be accounted for (McDaniel & Yoder, 2016). To account for these individual differences in internal and external factors contributing to lexical development in intervention planning, speech-language pathologists may benefit from looking

beyond the children's strengths and weaknesses in speech and language skills alone to those in other developmental domains that are interacting with their language skills (McDaniel & Yoder, 2016). Chapter 5 shows this 'whole person' perspective within a multiple case-study design, based on the ICF-CY and the body structures, body functions, activity and participation domains and the environmental factors playing a role in lexical development and communication. The Down syndrome behavioural phenotype, as described in the introduction, could not account for the individual differences in these ICF-CY domains, although children shared vocabulary sizes and/or mental ages. This shows the value of also conducting in-depth case-studies, next to group studies.

## A communicative perspective on lexical development and functional word use

Without sufficient levels of vocabulary knowledge, communication would suffer, since conveying a message would need at least a basic knowledge of words (Agdam & Sadeghi, 2014). Given this social nature of language development, the study of lexical development needs a communicative perspective, as is shown by several internal and environmental predictors of lexical development in Down syndrome in Chapters 4 and 5. Communication skills are associated with life opportunities (Cuskelly, Povey, & Jobling, 2016). Children with Down syndrome with strong communication performance skills, such as the level of communicative intent and expressive vocabulary size (see Chapter 5), are more likely to have a larger social network and keep friends (Wadman, Durkin, & Conti-Ramsden, 2011), to be in higher levels of education or employment (Foley et al., 2013), and to better acquire other skills such as reading (Nash & Heath, 2011). The level of vocabulary therefore has a substantial influence on the quality of life of individuals with Down syndrome (Cuskelly et al., 2016) and on the level of participation. This communicative perspective on word learning thus needs to be included in language theory.

Models of language generation and comprehension are often limited to fluent speech (Clark & Fox Tree, 2002), but human communication is multimodal in nature as people mostly use a combination of speech and non-speech modes to communicate and is highly influenced by the communicative context (Alant, Bornman, & Lloyd, 2006). Multiple models exist that describe processes during the generation of fluent speech, of which Levelt's '*The blueprint of the speaker* is one of the most influential models (Levelt, 1989, 1993, 1999). This language production model contains several components: (a) the conceptualizer, (b) the formulator, (c) the articulator, (d) the acoustic-phonetic processor, and (e) the speech comprehension system. According to Levelt (1994), each of these

components has its own processing resources, which ‘makes it possible that they can function in a modular, automatic fashion and in parallel. The speaker’s attention is largely spent on conceptual preparation; all the rest comes for free’ (p.18). Skilled language use involves a high degree of this language planning/ formulation automaticity. The user’s attention can thus usually be limited to planning and interpretation of a message.

In most, even close to all, children with Down syndrome, language planning, production and comprehension is not automatized, due to which attention must be spent on all components. Attention regulation is a necessary and sufficient condition for working and long-term memory storage and retrieval (for an overview of studies, see Kurland, 2011). If due to a lack of automatisation much attention is needed to complete grammatical or phonological encoding, fewer resources remain for other processes, particularly monitoring and articulation (Van Zaalen-op ‘t Hof, 2009). This closely relates to the common problems with speech intelligibility in children with Down syndrome (Kumin, 1994). Increased attention demands are found to negatively affect aspects of auditory comprehension (Murray, Holland, & Beeson, 1997) and spoken vocabulary (Hula, McNeil & Sung, 2007) in typically developing children. This is further reflected in the results of the present thesis. Chapter 4 showed attention levels to be predictive of the spoken lexical development of young children with Down syndrome. Chapter 5 showed the several functions of attention (f.e., joint attention with the communication partner, sustained attention on the task, or shifting attention from communication partner to a referent) relating to communicative and lexical development in these children.

Children with Down syndrome often experience speech problems. Although the exact nature and origin of speech problems associated with Down syndrome still have to be determined, it is generally accepted that these speech problems often lead to significantly decreased intelligibility (Kumin, 2006) and problems in fluency development. The speech errors of children with DS do not merely reflect delayed acquisition. More than half the words of Down syndrome children are pronounced differently on repeated productions (Dodd & Thompson, 2001), where typically developing children produce less than 10% of the same words inconsistently (Burt, Holm, & Dodd, 1999), and children with delayed phonological development have an inconsistency rating less than 20% (Dodd, 1995). In the present thesis, inconsistencies in production were not taken into account in determining vocabulary size. Dodd and Thompson (2001) found that children with Down syndrome make fewer errors in imitation than in spontaneous production of the same words (Dodd & Thompson, 2001). These findings show problems in both the segmental (i.e., syllables) and metrical spellout in children

with Down syndrome (for the adaptation of Levelt's phonological encoding, see Nijland, 2003).

Speech difficulties in individuals with Down syndrome are also often characterized as a result of: (1) a specific physiological and anatomical profile, including a smaller than average oral cavity, hypotonia of muscles around the mouth, fusion of lip muscles, and extra lip musculature (Miller, Leddy, & Leavitt, 1999), and additional problems in speech motor control (Kumin, 1994); (2) a phonological delay (Stoel-Gammon, 2001; Van Borsel, 1996); (3) signs of dyspraxia (Kumin, 2006), and (4) hearing loss (Dodd & Thompson, 2001). These findings indicate that there may be problems at all levels of Levelt's model. Van Zaalen and Reichel (2015) show that Levelt's model is very useful as an explanatory model to indicate the causes of production problems. A production problem may be related to the conceptualizer (f.e., low levels of communicative intent), grammatical encoding, phonological encoding (segmental and metrical), motor planning or programming and/or execution. Given this explanatory power, Van Zaalen and Reichel state that this model should be incorporated both in language and communication theory and clinical-decision making in these fields.

Next to speech, as the present thesis showed, manual sign use is an important mode of communication in young children with Down syndrome and a means to support receptive and expressive vocabulary development. Levelt's model was based entirely on speech output, not showing the multimodality in utterances that can be seen in children with Down syndrome. Given that word learning and functional word use is dependent on the communicative setting, context and partner, Levelt's model can benefit from taking this communicative perspective. The model is frequently used and adjustments have been made within psycholinguistics and other related fields that study language and communication development. For example, it has been adapted to help to understand the relationship between speech and gestures within aphasic patients within the sketch model (De Ruiter, 2000) and the interface model (Kita & Özyürek, 2003).

The sketch model (De Ruiter, 2000) clearly shows that communicative intent can be conveyed by a gesture and thus by other communication modes than speech. Especially for individuals that are not (yet) able to speak (intelligibly), as are many young children with Down syndrome, this indicates that non-speakers are communicators as well, and are trying to convey communicative intent. The contribution of the interface model of Kita and Özyürek (2003) mostly lies in the "empirically well-motivated elaboration of processes assumed to take place in the conceptualiser in the Sketch Model to accommodate cross-linguistic results" (De Ruiter & De Beer, 2013). Kita and Özyürek formulated a speech and gesture production model in which a negotiation exists within a communication planner

between a message generator (i.e. the speech path) and the action generator (i.e., the gesture path) to ascertain that the resulting speech and gesture express similar information. The communication planner, that links to Levelt's conceptualizer, decides what modalities of expression should be involved, based on the discourse model and environment (Kita & Özyürek, 2003). Given that children with Down syndrome are multimodal communicators (see Chapters 2, 3, and 4), a multimodal 'pathway' from conceptualizer to articulator should be distinguished within the model. Although the communication planner in the interface model might provide this opportunity, only speech is directly linked to the lexicon. The young children with Down syndrome in the longitudinal study (see Chapter 4) had a large amount of manual signs in their lexicon at the start of the study ($M$ = 85; *range*: 0 – 301 words), showing that the lexicon should be linked to multimodal processing and output.

Although these models discuss important contributions to Levelt's model, speech is still seen as the most important communication mode. This issue has been taken into account in the adaptation of Emmorey, Borinstein, Thompson, and Gollan (2008) from the perspective of bimodal bilingualism in which individuals are fluent in spoken and sign language. Separate perception and motor systems provide these individuals with the opportunity to produce and perceive two languages in different modalities at the same time, with the (simultaneous) use of speech and signs (Emmorey et al., 2008). The most important contribution from this model, elaborating on the action generator, is that a message can be formulated in different communication modes via manual articulation (sign or gesture) and/or vocal/facial articulation (speech or non-manual sign language components), which is highly relevant for Down syndrome. Emmorey's model was tested with eleven healthy, fluent bimodal bilinguals who are able to code-switch immediately between speech and manual signs. The American Sign Language (ASL) and English Formulators are hypothesised to contain the lexicons and grammatical, morphological, and phonological encoding processes for each language (Emmorey et al., 2008). Emmorey et al. assumed these are distinct for sign language and spoken language. Young children with Down syndrome are neither fluent in speech, nor in manual signing or in other modes of communication. Also, the children with Down syndrome in the present studies were not learning sign language, but make use of Sign Supported Dutch, in which manual signing is (more) closely linked to spoken language than in sign language. As can be seen in the data from Chapters 2 and 3, young children with Down syndrome are able to produce some words only in speech, some words only in manual signs, and some words in both modalities. This may indicate a shared lexicon in which both modalities can be stored, rather than two separate pathways.

Chapter 4 and 5 show the role of internal and external factors in the communicative and lexical development of children with Down syndrome. Several of these internal factors can be found in abovementioned models, such as attention, working memory, motor development, the level of communicative intent, and phonological and phonemic awareness. The role of external factors, factors within the child's (in)direct environment (f.e., communication partners), have not been distinguished within earlier models. Although Kita and Özyürek (2003) and Emmorey et al. (2008) included a communication planner, all these models only provide a pathway for the sender of a message. Except for the original model of Levelt (1993), none of the adaptations included a comprehension system, secluding the possibility to portray language and vocabulary use within a communicative perspective. Based on the results of the studies in this thesis and all previously described models, a theoretically and practically driven model is proposed which I will call '*the blueprint of the multimodal communicator*', after Levelt's 'blueprint of the speaker', which integrates the communicative perspective in lexical development (see Figure 1, p. 137).

In *the blueprint of the multimodal communicator*, the following adjustments to Levelt's original model (1993) are proposed:

(a) Including the communication planner from Kita and Özyürek (2003) as an adjustment of the conceptualizer. The communication planner, based on environmental input, discourse model, and communicative intent, decides what modalities of expression should be involved and whether the action generator and/or the formulator should be activated. Communicative intent was found in the present thesis as a predictor of expressive vocabulary development and communication performance in children with Down syndrome (see Chapter 4 and 5).

(b) Including the action generator from Kita and Özyürek (2003), since a communicative intent can be conveyed by just pointing towards or grabbing a referent, for example with the communicative intent 'to want'. In the event that the generation of either the preverbal message or the gesture is hampered, theconceptualizer will compensate by shifting a higher load to the other channel (De Ruiter, 2000).

(c) The mental lexicon is adjusted to a multimodal mental lexicon to include vocabulary in other modalities than only speech, since the present thesis showed that the lexicon of young children with Down syndrome is multimodal in nature. For children with Down syndrome this multimodal lexicon in influenced by both internal and external factors (see Chapters 4 and 5).

(d) Within the phonological encoding, I included the additions of Nijland (2003) in order to be able to investigate possible causes of unintelligible speech,

common in children with Down syndrome, on the level of phonological encoding. Although this is not a direct finding of the present thesis, these additions are important to construct a comprehensive model of multimodal language production and communication.

(e) Given the multimodality of the lexicon and of communication of children with Down syndrome, a motor plan is added as result of the formulator, next to a phonetic plan, if the message is conveyed via other communication modes, such as manual signing.

(f) The articulator only leading to speech output is adjusted to a multimodal articulator with output opportunities in several modalities and communication modes, such as speech, manual signs, gestures, pointing, grasping, eye gaze and/or body language.

(g) A comprehension system of both communication partners, and not only as part of the self-monitoring system, related to the multimodal mental lexicon. Communication infers that there is a sender and a receiver, who interchangeably may take either role (Van Balkom & Welle Donker – Gimbrère, 2004). Based on the previous utterance or actions of the sender, the receiver may respond, first conceptualizing the idea to utter himself, leading him through the same processes. Given the communicative perspective, the model should be seen as a loop or continuous process in which the utterance of the first will elicit the communication planner of the second to start generating a message. The language comprehension system comprises the following two component processes (Levelt, 1992): (I) Perceptual decoding: mapping input onto some code that is linguistically parsed, which involves segmenting and recognizing words (phonological and morphological decoding, and accessing the multimodal mental lexicon), as well as assigning syntactic and semantic structure, (II): Interpreting: "inferring the intended meaning by identifying referents and computing a conceptual representation for the utterance on the basis of the result of perceptual decoding, along with prosody and contextual information" (p. 291).

(h) A multimodal monitoring system, including self-and other monitoring, where Levelt only described self-monitoring. In communication, people also rely on *other-monitoring*, closely linked to processes of perception, cognition and socio-emotional development as described in chapter 5. Communicators monitor not just their own actions and monitor whether the (c)overt messages match the communicative intent, but also those of their communication partners, taking both into account as they produce utterances (Clark & Krych, 2004). Communication partners try to keep each other informed of their current state of understanding. They try to ground what has been intended; make it part of their common ground (Clark & Schaefer, 1987). Either negative or positive 'evidence'

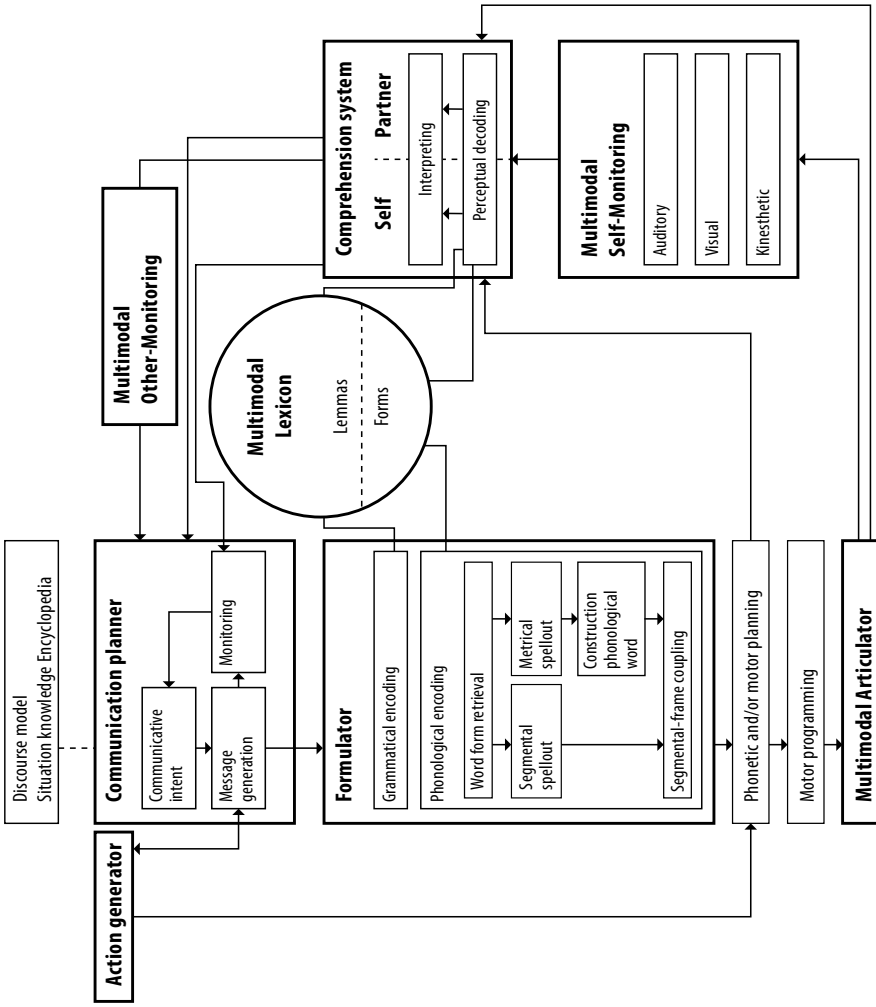



Figure 1. *The blueprint of the multimodal communicator.*

in the reactions of the communication partner to the utterance may serve as a feedback mechanism (Clark & Brennan, 1991). Negative evidence shows that the person has been misheard or misunderstood; the communicative intent was not shared and no common ground was established. The person, if in good health and communicating with a known relative, congruently will try to repair the problem by reformulating the utterance, perhaps making use of different words from the lexicon. Positive evidence includes continuers such as *uh huh* and *yes* or *yeah* or an initiation of the 'relevant or appropriate next turn' (e.g., the answer to a question, the response to a request, the acceptance of an invitation by the communication partner (Clark & Brennan, 1991). This other-monitoring, as is the case with self-monitoring, not only relies on the monitoring of speech, as is only the case in Levelt's model. Next to auditory monitoring, it needs *multimodal monitoring* (e.g., visual, kinesthetic) in order to also monitor manual signs, body language, eye gaze, gestures, etcetera. Fluent communicators may directly reformulate their message to establish comprehension with their communication partner. For young children with Down syndrome, this will be much harder to accomplish, especially with the use of manual signs and their unintelligible speech, to which not every communication partner may be acquainted. Children with Down syndrome show impairments in the phonological loop of working memory, which feed into the planning and articulation of meaningful words (Jarrold & Baddeley, 2001). Chapter 4 also showed the level of phonological and phonemic awareness to be highly predictive of the number of spoken words of young children with Down syndrome. Manual signs might be a more accessible communicative tool for children with Down syndrome (Özçaliskan et al., 2016). Reformulating a message may ask for a code-switch between speech and manual signing, if the child's communication partner did not understand the spoken utterances, or by selecting synonyms from the lexicon. The success in establishing common ground for these children with Down syndrome and using words from their mental lexicon in a functional way is therefore highly relying on the communicative context and communication partner.

## Future directions

The studies presented in Chapters 2, 3, 4 and 5 provided important insights in the lexical development of children with Down syndrome. Each of these studies, however, had its limitation or leaves us with further questions, that should be accounted for in future research: (a) the sample size, (b) only vocabulary size (i.e., breadth of vocabulary) was taken into account, and (c) the functional words use (i.e., core vocabulary) should be investigated in more depth.

Firstly, although the recruitment of participants with Down syndrome is not easy and small sample sizes are common in cross-sectional and longitudinal studies with children with Down syndrome, this restriction limits the generalisability of the findings of this study. The total model of longitudinal predictors of lexical development (see Chapter 4) could therefore not be assessed on goodness-of-fit due to a lack of power given the relatively small sample size (N = 20). A structural relations model of the expressive and receptive vocabulary development, based on successive stepwise multilinear regressions, was presented. In a future study, this structural relations model of lexical development should be tested in a larger population with a Structural Equation Modeling (SEM) technique, both in children with Down syndrome as well as in a control group of typically developing children. SEM is a statistical approach to model means and covariances among multivariate data and allows for a simultaneous analysis of all variables in the model, visualized by a graphical path diagram. The small sample size in Chapter 5 did allow for an in-depth 'whole person' view on the functioning of the children. Such a multiple-case study can disentangle all internal and external factors linked to communicative and lexical development and is highly informative for clinical practice in the field of assessment and clinical-decision making. Next to identifying predictors on the group level, more in-depth assessment of lexical development is needed to show individual strengths and weaknesses.

Secondly, most studies describing the lexical development of children with Down syndrome, including the present thesis, focus on the breadth of vocabulary. Vocabulary breadth can be seen as the number of phonological entries within the lexicon that can be mapped to the correct semantic representations (Christ, 2011). To date, less attention is given to the depth of their vocabularies: the extent to which word meanings have been refined and semantic knowledge has been elaborated (Christ, 2011), also called 'deep word knowledge'. Wesche and Paribakht (1996) defined deep word knowledge as the richness of the representation of the known words or how well a child knows about words or concepts, which closely links to the comprehension system and especially 'interpreting' in the blueprint of the communicator. Typically developing children build their lexicon in ways such that learning new words is dependent on semantic relations among words already acquired or semantic relations in the learning environment (Hills et al., 2009). When a child increases its vocabulary size, the new words to be learned need to be related and attached to a network of already know words (Agdam & Sadeghi, 2014). The more detailed the semantic knowledge the more efficient lexical access will be and the less working memory load is needed to retrieve words from and store words in the lexicon (Bjorklund, 1987). When semantic knowledge is more detailed, new words will rapidly form connections with already learned words

within the lexicon. Children at risk for significant language delay merely add words as individual and unrelated items, not picking up on the semantic relations in the learning environment (Beckage, Smith, & Hills, 2010).

A study in children with Specific Language Impairment (SLI; Sheng & McGregor, 2010) found a deficit in the organization of the lexical-semantic system in these children, especially for those children with a deficit in expressive vocabulary and a gap between receptive and expressive vocabulary, also often seen in children with DS. SLI children mostly gave sound-based connections between items in the lexicon, which suggest fewer or less-robust semantic links than in typically developing children. For children with Down Syndrome and children with intellectual disabilities (ID) in general, little is known about the development of deep lexical knowledge and the capacities to build and retrieve meaning associations. Where fast mapping experiments suggest that individuals with DS learn new vocabulary as readily as mental age-matched typically developing children (f.e. Kay-Raining Bird et al., 2004), these do not provide insight in semantic organization of these new learned words. Deckers and Stoep (2014) conducted a pilot study, using a receptive and expressive word association task, in six children with Down syndrome between the age of 6;0 and 8;0 years, matched on expressive vocabulary size with six typically developing children. These preliminary results showed that the semantic knowledge of the children with Down syndrome closely resembled that of their peers, although the children with Down syndrome could name fewer associations with a referent. Instructional strategies aimed at the child's lexical development may be even more effective when these are in accordance with the development of deep lexical knowledge. In order to gain more insight in the semantic representation of the lexical of children with Down syndrome, research into the assessment of deep lexical knowledge in children with Down syndrome is needed.

Thirdly, the study towards core vocabulary of children with Down syndrome (see Chapter 3) is the first study in children with intellectual disabilities with a mental age below 4 years (for a review of core vocabulary studies in (a) typically developing populations, see Van Tilborg & Deckers, 2016). Given that AAC support teams and SLPs rarely have enough knowledge and experience to select vocabulary for functional use for specific contexts, activities, ethnicity, or language group (Beukelman & Mirenda, 2013), future research has to focus on the functional language use of groups from different (a)typical populations. Although providing non-speaking or unintelligible children with the multimodal vocabulary needed for communication is of highest importance, to date, no other core vocabulary studies have been conducted in Dutch, typically or atypically developing children, besides the study described in Chapter 3. Core words can

maximize the potential for spontaneous language generation, in spoken, signed, graphic and/or written (output) modalities. Incorporating this core vocabulary into language interventions and/or in an AAC system may provide users the opportunity to be engaged in communication and interaction in an appropriate, efficient and relatively quick manner (Hill, Kovacs, & Shin, 2015; Weighton & Dodd, 2011). This asks for future studies investigating the core vocabulary of several (Dutch) atypical populations, such as autism spectrum disorders (ASD), specific language impairments (SLI), and other syndromes related to language and communication problems like Down syndrome. Since communication is not limited to home and therapy sessions, language samples should be collected in more settings and with different communication partners. Future studies on core vocabulary should include other contexts as well, such as the (pre)school classroom, and activities with siblings or peers, in order to also investigate the commonality of vocabulary use over a range of contexts and communication partners. And the effect of incorporating these words, combined with content words (i.e., fringe vocabulary), during language intervention on the lexical development and communicative performance of these children should be focused on.

## Clinical implications



Significantly delayed onset of speech and poor intelligibility are two primary reasons for introducing Augmentative and Alternative Communication (AAC) to children with Down syndrome (Brady, 2008). The primary purpose of AAC is to increase opportunities for communication by providing an additional modality or by enforcing, supporting existing modalities through which individuals can enhance their communication with a variety of different people in their lives, such as parents, siblings, peers, and educators (Barker, Akaba, Brady, & Thiemann-Bourque, 2013). If children with Down syndrome are able to understand more than they are able to produce and if there is a way to bypass their speech production problem, they should become more effective communicators (Kay-Raining Bird et al., 2000; Kumin, 2003; Miller, 1999). Given that language learning takes place through communication and, as has been found in the present thesis in children with Down syndrome, is propelled by the communicative intent of a child (Kühn & Langner, 2012), providing different means for communication, such as manual signs, positively influences lexical development. Making words more perceptually salient by producing them in single-word utterances or final utterance position (Fernald & Mazzie, 1991), reducing the complexity of the utterance, providing clear cues as to the referent for a word (Hoff & Naigles, 2002), and using multi-

modal input all positively impacts novel word learning in children with Down syndrome (Kay-Raining Bird & Cleave, 2015).

Lexical development for children using AAC is also influenced by how adults model or shape the use of this augmented language (Romski & Sevcik, 2003), as is the case with learning to speak as well, which again shows the need for a communicative perspective on vocabulary learning. This augmented input supports the development by providing models of AAC usage, illustrating the usefulness, and demonstrating that it is an acceptable way of communicating. Augmented input illustrates the real-world meaning of symbols and signs, the many functions that they can serve, and it demonstrates that AAC is both accepted and encouraged as a modality for communication (Sevcik & Romski, 2002). Augmented, multimodal input is therefore recognized as an important ingredient for children to learn new vocabulary and to increase expressive and receptive language skills (Barker et al., 2013). In 6- to 8-year old children with Down syndrome, vocabulary levels were a strong predictor of social behaviour (Næss, Nygaard, Ostad, Dolva & Halaas Lyster, 2016). Receptive vocabulary predicted social problems, while expressive vocabulary predicted social capabilities. Næss and colleagues conclude that because vocabulary emerged as an important predictor of social functioning, lexical development should be the focus of early interventions for children with Down syndrome.

When children use multimodal forms of communication, as do most young children with Down syndrome, this has to be accounted for in the assessment of lexical development. The present thesis showed that the N-CDI is a valid measurement of multimodal vocabulary. The adapted version (see Chapter 2) can easily be used in clinical practice and is a cost- and time-effective way to gain insight in the size of the vocabulary of young children with Down syndrome. Next to the N-CDI, the functional use of words should be identified to gain insight in the way that children use their multimodal vocabulary in everyday communication. Analyses relating to determining core vocabulary (see Chapter 3) can be used as assessment method in clinical practice as well. Language sample collection and analysis is an historically used evidence-based practice for selecting vocabulary for speech-language therapy and AAC interventions (Kovacs & Hill, 2015). Vocabulary selection should be guided by two main principles: (a) the need to convey essential messages and (b) the eventual development of language skills (Beukelman & Mirenda, 2013). Selected vocabulary should be highly supportive of language and communication development because vocabulary development is key in language learning (Hohenberger & Peltzer-Karpf, 2009). Within the assessment of lexical development focus should be on both core and fringe vocabulary.

The present thesis has shown that the lexical development needs a communicative perspective. Professionals working with and parents of children with Down syndrome should become aware of all predictors and processes influencing the lexical development of these children. In speech-language therapy and AAC interventions, all these predictors and processes should be assessed and taken into account in clinical-decision making. Language and communication require interprofessional collaboration, since the present study showed that it is about more than only vocabulary and speech. In interprofessional collaboration several involved disciplines, in close collaboration with the child and/or family, formulate shared intervention goals, speak the same language, which is accessible and understandable for all involved, and perceive the qualities and perspectives of other disciplines as complementary and valuable (Buntinx & Bijwaard, 2004). Given the number and variety of internal and external predictors of lexical development, as identified in the present thesis, multiple disciplines (e.g., speech-language pathology, occupational and physical therapy, teachers, psychology/ behavioural and educational science) have to work together more closely. Involved professionals have to communicate more openly, both with other professionals as well as with parents. The ICF-CY provides a useful framework and the shared language needed for clinical practice to construct an integrative profile of lexical development and communicative performance, including body functions, structures, activities and participation, and environmental and personal factors (see Chapter 5). Intervention planning in children with Down syndrome should include this 'whole person' perspective. Professionals have to look beyond the borders of their own discipline and should integrate insights from colleagues into their clinical practice in order to create the best learning environments for children with Down syndrome to learn new words. Within a communicative perspective on lexical development, all communication partners in the social network of the child with Down syndrome should be involved.

# Chapter 7

## References

# References

Abbeduto, L., Murphy, M. M., Cawthon, S. W., Richmond, E. K., Weissman, M. D., Karadottir, S., & O'Brien, A. (2003). Receptive language skills of adolescents and young adults with Down or fragile X syndrome.American Journal on Mental Retardation,108(3), 149-160.

Abbeduto, L., Pavetto, M., Kesin, E., Weissman, M., Karadottir, S., O'Brien, A., & Cawthon, S. (2001). The linguistic and cognitive profile of Down syndrome: Evidence from a comparison with fragile X syndrome. Down Syndrome Research and Practice, 7(1), 9-15. DOI:10.3104/reports.109

Abbeduto, L., Warren, S. F., & Conners, F. A. (2007). Language development in Down syndrome: From the prelinguistic period to the acquisition of literacy. Mental Retardation and Developmental Disabilities Research Reviews, 13(3), 247-261.

Achenbach, T. M., & Rescorla, L. (2000). ASEBA preschool forms & profiles: An integrated system of multi-informant assessment. Aseba.

Adamson, L. B., Bakeman, R., & Brandon, B. (2015). How parents introduce new words to young children: The influence of development and developmental disorders. Infant Behavior and Development, 39, 148-158. doi:10.1016/j.infbeh.2015.02.008.

Adamson, L. B., Romski, M., Deffebach, K., & Sevcik, R. A. (1992). Symbol vocabulary and the focus of conversations: Augmenting language development for youth with mental retardation. Journal of Speech and Hearing Research, 35, 1333 – 1343. doi:10.1044/jshr.3506.1333

Adolfsson, M. (2011). Applying the ICF-CY to identify everyday life situations of children and youth with disabilities. Unpublished PhD thesis, Jönköping, Sweden.

Agdam, S. J., & Sadeghi, K. (2014). Two formats of word association tasks: A study of depth of word knowledge. English Language Teaching, 7(10), 1-12.

Aitchison, J. (2003). Words in the mind. An introduction to the mental lexicon. Maiden, MA: Blackwell.

Akhtar, N., & Tomasello, M. (1998). Intersubjectivity in early language learning and use. In S. Braten. Intersubjective communication and emotion in early ontogeny (pp. 316-335). Cambridge: Cambridge University Press.

Akhtar, N., & Tomasello, M. (2000). The social nature of words and word learning. In R. M. Golinkoff et al. (eds). Becoming a word learner: A debate on lexical acquisition (pp. 115-135). Oxford: Oxford University Press.

Alant, E., Bornman, J., & Lloyd, L. L. (2006). Issues in AAC research: How much do we really understand?. Disability and rehabilitation, 28(3), 143-150.

American Association on Intellectual and Developmental Disabilities (2016). Definition of intellectual Disability. https://aaidd.org/intellectual-disability/definition#.VuLJqxFVhBc

Anderson, V., Anderson, P. J., Jacobs, R., & Smith, M. S. (2008). Development and assessment of executive function: From preschool to adolescence. In V. Anderson, R. Jacobs, and P. J. Anderson. Executive functions and the frontal lobes: A lifespan perspective (pp. 123-154). New York: Psychology Press.

Anderson, P. J., De Luca, C. R., Hutchinson, E., Roberts, G., & Doyle, L. W. (2010). Underestimation of developmental delay by the new Bayley-III Scale. Archives of Pediatrics & Adolescent Medicine, 164(4), 352-356.

Anderson, V., Northam, E., Hendy, J., & Wrennall, J. (2001). Developmental neuropsychology: a clinical approach. New York: Psychology Press.

Anderson, D., & Reilly, J. (2002). The MacArthur communicative development inventory: normative data for American Sign Language.Journal of Deaf Studies and Deaf Education, 7(2), 83-106.

Attwood, A., Frith, U., & Hermelin, B. (1988). The understanding and use of interpersonal gestures by Autistic and Down's syndrome children. Journal of Autism and Developmental Disorders, 18 (2), 241-257.

Baddeley, A. (2012). Working memory: theories, models, and controversies. Annual Review of Psychology, 63, 1-29.

Bahrick, L. E., & Lickliter, R. (2002). Intersensory redundancy guides early perceptual and cognitive development. Advances in child development and behavior, 30, 153-189.

Baker, B., Hill, K., & Devylder, R. (2000, March). Core vocabulary is the same across environments. Paper presented at the California State University at Northridge Conference, Los Angeles, CA. Abstract retrieved from http://minspeak.com/students/documents/2000CSUN.pdf.

Balandin, S., & Iacono, T. (1999). Crews,wusses, and whoppas: Core and fringe vocabularies of Australian meal-break conversations in the workplace. Augmentative and Alternative Communication, 15, 95–109. doi:10.1080/07434619912331278605 doi:10.1080/07434619912331278605 doi:10.1080/07434619912331278605

Banajee, M., Dicarlo, C., & Buras Stricklin, S. (2003). Core vocabulary determination for toddlers. Augmentative and Alternative Communication, 19, 67–73. doi:10.1080/0743461031000112034

Bandura, A. (1969). Principles of behavior modification. New York, Holt, Rinehart & Winston.

Barker, R. M., Akaba, S., Brady, N. C., & Thiemann-Bourque, K. (2013). Support for AAC use in preschool, and growth in language skills, for young children with developmental disabilities. Augmentative and Alternative Communication, 29(4), 334-346.

Bates, E., Camaioni, L., & Volterra, V. (1975). The acquisition of performatives prior to speech. Merrill-Palmer Quarterly of Behavior and Development, 21(3), 205-226.

Beckage, N., Smith, L., & Hills, T. (2010). Semantic network connectivity is related to vocabulary growth rate in children. The Annual Meeting of the Cognitive Science Society, 2769-2774.

Behrens, M., & Hauch, J. (2011). Does motor development influence language development? Poster presented at the CECP GSO Research Exchange Conference.

Beitchman, J. H., Jiang, H., Koyama, E., Johnson, C. J., Escobar, M., Atkinson, L., Brownlie, E. B., & Vida, R. (2008). Models and determinants of vocabulary growth from kindergarten to adulthood. Journal of Child Psychology and Psychiatry, 49(6), 626-634.

Bello, A., Onofrio, D., & Caselli, M. C. (2014). Nouns and predicates comprehension and production in children with Down syndrome.Research in Developmental Disabilities, 35(4), 761-775.

Berglund, E., Eriksson, M., & Johansson, I. (2001). Parental reports of spoken language skills in children with Down syndrome. Journal of Speech, Language and Hearing Research, 44, 179-191.

Beukelman, D., Jones, R., & Rowan, M. (1989). Frequency of word usage by nondisabled peers in integrated preschool classrooms.Augmentative and Alternative Communication, 5, 243 - 248. doi: 10.1080/07434618912331275296

Beukelman, D., & Mirenda, P. (2013). Augmentative and alternative communication: Supporting children and adults with complex communication needs (4th ed.). Baltimore, MD: Paul H. Brookes.

Bjorklund, D. F. (1987). How age changes in knowledge base contribute to the development of children's memory: An interpretive review. Developmental Review, 7(2), 93-130.

Bloom, P. (2000). How children learn the meanings of words. Cambridge, MA: MIT press.

Blackstone, S. & Hunt Berg, M. (2003). Social Networks Inventory : A communication inventory for individuals with Complex Communication Needs and their Communication Partners. Monterey: Augmentative Communication Inc.

Boenisch, J. (2014). Kernvokabular im Kindes- und Jugendalter. UK & Forschung, 3, 4–23.

Boenisch, J., & Soto, G. (2015). The oral core vocabulary of typically developing English-speaking school-aged children: Implications for AAC practice. Augmentative and Alternative Communication, 31, 77–84. doi:10.3109/07434618.2014.1001521

Boenisch, J., & Sachse, S. (2007). Zum Einsats von Kern- in der frühen Förderung [The use of core vocabulary in the early development]. Unterstützte Kommunikation, 3, 12–20.

Bolier, W. (2010). Communicatie in Nederlands met Gebaren in het speciaal onderwijs aan dove, slechthorende en ESM-kinderen [Communication in Sign Supported Dutch in special eduction for deaf, hard of hearing children and children with specific language impairments]. VHZ Artikelen, 6, 14-18.

Bol, G. & Kuiken, F. (1989). Handleiding GRAMAT. Methode voor het diagnosticeren en kwalificeren van taalontwikkelingsstoornissen [Instruction manual GRAMAT. Method for diagnostics and qualifying specific language impairments]. Lisse: Swets B.V.

Bonvillian, J. D. (2002). Lending a helping hand: The development of a simplified sign system. Invited address, 44th annual conference of the Speech-Language-Hearing Association of Virginia, Charlottesville, VA.

Borghi, A. M., & Cimatti, F. (2010). Embodied cognition and beyond: Acting and sensing the body. Neuropsychologia,48(3), 763-773.

Bornstein, M. H., & Putnick, D. L. (2012). Stability of language in childhood: A multiage, multidomain, multimeasure, and multisource study.Developmental Psychology, 48(2), 477.

Borsel, J. V. (1996). Articulation in Down's syndrome adolescents and adults. European Journal of Disorders of Communication, 31(4), 415-444.

Bowey, J. A. (2001). Nonword repetition and young children's receptive vocabulary: A longitudinal study.Applied Psycholinguistics, 22(03), 441-469.

Brady, N. C. (2008). AAC for children with Down syndrome and children with fragile X syndrome. In: Roberts, J. E., Chapman, R. S., Warren, S. F. (eds.). Speech and language development and intervention in Down syndrome and fragile X syndrome (pp. 255–274). Baltimore: Brookes.

Brady, N. C., Marquis, J., Fleming, K., & McLean, L. (2004). Prelinguistic predictors of language growth in children with developmental disabilities. Journal of Speech, Language, and Hearing Research, 47(3), 663-677.

Brady, N. C., Thiemann-Bourque, K., Fleming, K., & Matthews, K. (2013). Predicting language outcomes for children learning augmentative and alternative communication: Child and environmental factors.Journal of Speech, Language, and Hearing Research, 56(5), 1595-1612.

Brent, M.R., & Siskind, J.M. (2001). The role of exposure to isolated words in early vocabulary development. Cognition, 81, B33–B44.

Broese van Groenou, M.I., & Deeg, D.J.H. (2006). Veranderingen in sociale participatie. In: A. de Boer (red.). Rapportage Ouderen (pp. 215-238). Den Haag: SCP.

Broshek, D. K., & Barth, J. T. (2000). The Halstead-Reitan neuropsychological test battery. In G. Groth-Marnat (Ed). Neuropsychological assessment in clinical practice: A guide to test interpretation and integration (pp. 223-262). Hoboken: John Wiley & Sons.

Brown, J. H., Johnson, M. H., Paterson, S. J., Gilmore, R., Longhi, E., & Karmiloff-Smith, A. (2003). Spatial representation and attention in toddlers with Williams syndrome and Down syndrome. Neuropsychologia, 41(8), 1037-1046.

Brownell, R. (2000). Expressive One-Word Picture Vocabulary Test. Novato, CA: Academic Therapy.

Brownell, R. (2000). Receptive One-Word Picture Vocabulary Test. Novato, CA: Academic Therapy.

Buckley, S., & Bird, G. (2009). Memory development for individuals with Down syndrome– An overview. Cumbria, UK: Down Syndrome Education International.

Buckley, S., Bird, G., & Sacks, B. (2002). Social development for individuals with Down syndrome-An overview. Cumbria, UK: Down Syndrome Education International.

Buntinx, W. H. E, & Bijwaard, M. (2004). Professionaliteit in de zorg voor mensen met functiebeperkingen – Kenmerken, rol, voorwaarden. Utrecht: Vereniging Gehandicaptenzorg Nederland.

Burt, A., Holm, B., & Dodd, L. (1999). Phonological awareness skills of 4-year-old British children: An assessment and developmental data. International Journal of Language & Communication Disorders, 34(3), 311-335.

Butterworth, G. (2003). Pointing is the royal road to language for babies. In S. Kita, Pointing: Where language, culture, and cognition meet (pp 9-33). Hillsdale, NJ: Erlbaum.

Calandrella, A. M., & Wilcox, M. J. (2000). Predicting language outcomes for young prelinguistic children with developmental delay. Journal of Speech, Language, and Hearing Research, 43(5), 1061-1071.

Campbell, W. N., & Skarakis-Doyle, E. (2007). School-aged children with SLI: The ICF as a framework for collaborative service delivery. Journal of Communication Disorders, 40(6), 513-535.

Capirci, O., Caselli, M. C., & De Angelis, V. (2010). Gesture for speaking and gesture for thinking in children with typical and atypical development: a new theoretical framework. In D. Riva and C. Njiokiktjien. Brain lesion localization and developmental functions: basal ganglia, connecting systems, cerebellum, mirror neurons (p. 201- 217). Montrouge: John Libbey Eurotext.

Capone, N. C. (2007). Tapping toddlers' evolving semantic representation via gesture. Journal of Speech, Language, and Hearing Research,50(3), 732-745.

Caselli, M. C., Vicari, S., Longobardi, E., Lami, L., Pizzoli, C., & Stella, G. (1998). Gestures and words in early development of children with Down syndrome. Journal of Speech, Language, and Hearing Research,41(5), 1125-1135.

Case-Smith, J. (2001). Occupational therapy for children. St. Louis, MO: Mosby. Centrum voor Consultatie en Expertise. (2010). Vragenlijst kwaliteit van bestaan [Questionnaire quality of living]. Utrecht: CCE.

Chan, J., & Iacono, T. (2001). Gesture and word production in children with Down syndrome. Augmentative and Alternative Communication, 17, 73-87. doi:10.1080/aac.17.2.73.87

Chapman, R.S. (1995). Language development in children and adolescents with Down syndrome. In P. Fletcher & B. MacWhinney (Eds.), Handbook of Child Language. (pp.651-663). Oxford: Blackwell.

Chapman, R. (2006). Language learning in Down syndrome: the speech and language profile compared to adolescents with cognitive impairment of unknown origin. Down Syndrome Research and Practice, 10(2), 61-66.

Chapman, R. S. & Hesketh, L. J. (2000). Behavioral phenotype of individuals with Down syndrome. Mental Retardation and Developmental Disabilities Research Reviews, 6, 84–95.

Chapman, R. S., Hesketh, L. J., & Kistler ,D. J. (2002). Predicting longitudinal change in language production and comprehension in individuals with Down syndrome: hierarchical linear modeling. Journal of Speech, Language, and Hearing Research, 45, 902–915.

Chapman & Kay-Raining Bird (2012). Language development in childhood, adolescence, and young adulthood in persons with Down syndrome. In J. A. Burack, R. M. Hodapp, G. Iarocci, & E. Zigler (Eds.). The Oxford Handbook of Intellectual Disability and Development (pp. 167– 183). Oxford: Oxford University Press.

Chapman, R. S., Schwartz, S. E., & Bird, E. K. R. (1991). Language skills of children and adolescents with Down Syndrome I. Comprehension. Journal of Speech, Language, and Hearing Research, 34(5), 1106-1120.

Chapman, R. S., Seung, H. K., Schwartz, S. E., & Kay-Raining Bird, E. (1998). Language skills of children and adolescents with Down syndrome II. Production deficits. Journal of Speech, Language, and Hearing Research, 41(4), 861-873.

Chapman, R. S., Seung, H. K., Schwartz, S. E., & Kay-Raining Bird, E. (2000). Predicting language production in children and adolescents with Down Syndrome. The role of Comprehension.Journal of Speech, Language, and Hearing Research, 43(2), 340-350.

Chen, M., Chen, Y., & Chen, C. (2013, August). Do they need specific language training content?: results of core vocabulary investigation from children with/without intellectual disabilities in

Taiwan. Paper presented at the Proceedings of the 7th International Convention on Rehabilitation Engineering and Assistive Technology, Singapore.

Chen, M. C., Hsu, S. C., Chi, P. H., Ko, C. C., Lin, Y. L., & Huang, Q. Y. (2011). Do they narrate in same vocabulary? The result of core vocabulary investigation from children with/without asperger syndrome in Taiwan. I-CREATe 2011 - International Convention on Rehabilitation Engineering and Assistive Technology, 57–60.

Chiat, S., & Roy, P. (2008). Early phonological and sociocognitive skills as predictors of later language and social communication outcomes. Journal of Child Psychology and Psychiatry, 49(6), 635-645.

Chomsky, Noam (1959). A review of B.F. Skinner's Verbal behavior. In Readings in the Psychology of Language, Prentice-Hall.

Christ, T. (2011). Moving past "right" or "wrong" toward a continuum of young children's semantic knowledge. Journal of Literacy Research, 43, 130-158.

Cieza, A., Brockow, T., Ewert, T., Amman, E., Kollerits, B., Chatterji, S., ... & Stucki, G. (2002). Linking health-status measurements to the international classification of functioning, disability and health. Journal of Rehabilitation Medicine, 34(5), 205-210.

Clark, H. H., & Brennan, S. A. (1991). Grounding in communication. In l. B. Resnick, J. M. Levine, & S. D. Teasley (Eds.). Perspectives on socially shared cognition (pp. 127-149). Washington: APA Books.

Clark, H. H., & Fox Tree, J. E. (2002). Using uh and um in spontaneous speaking. Cognition, 84(1), 73-111.

Clark, H. H., & Krych, M. A. (2004). Speaking while monitoring addressees for understanding. Journal of Memory and Language, 50(1), 62-81.

Clark, H. H., & Schaefer, E. F. (1987). Collaborating on contributions to conversations. Language and Cognitive Processes, 2(1), 19-41.

Cleland, J., Wood, S., Hardcastle, W., Wishart, J., & Timmins, C. (2010). Relationship between speech, oromotor, language and cognitive abilities in children with Down's syndrome. International Journal of Language & Communication Disorders, 45(1), 83-95.

Cogan, G. B., Thesen, T., Carlson, C., Doyle, W., Devinsky, O., & Pesaran, B. (2014). Sensory-motor transformations for speech occur bilaterally. Nature, 507(7490), 94.

Cornish, K., & Wilding, J. (2010). Attention, genes, and developmental disorders. Oxford: Oxford University Press.

Couzens, D., Haynes, M., & Cuskelly, M. (2012). Individual and environmental characteristics associated with cognitive development in Down syndrome: A longitudinal study. Journal of Applied Research in Intellectual Disabilities, 25(5), 396-413.

Cuskelly, M., Povey, J., & Jobling, A. (2016). Trajectories of development of receptive vocabulary in individuals with Down syndrome. Journal of Policy and Practice in Intellectual Disabilities, 13, 111-119.

Dale, P. S. & Goodman, J. C. (2005). Commonality and individual differences in vocabulary growth. In M. Tomasello & D. I. Slobin. Beyond nature-nurture: Essays in honor of Elizabeth Bates (pp. 41-78). Mahwah: Lawrence Erlbaum Associates.

Dalen, H. E., Nyquist, A., Saebu, M., Roe, C., & Bautz-Holter, E. (2013). Implementation of ICF in goal setting in rehabilitation of children with chronic disabilities at Beitostolen Healthsports Centre. Disability and rehabilitation, 35(3), 198-205.

Dark, L., & Balandin, S. (2007). Prediction and selection of vocabulary for two leisure activities. Augmentative and Alternative Communication, 23, 288–99. doi:10.1080/07434610601152140

Daunhauer, L. A., & Fidler, D. J. (2011). The Down syndrome behavioral phenotype: implications for practice and research in occupational therapy. Occupational Therapy in Health Care, 25(1), 7-25.

Deckers, S. R. J. M., & Stoep, J. (2014, July). Insights in vocabulary organization in children with Down Syndrome. Lecture presented at the ISAAC congress, Lisbon, Portugal.

Deckers, S. R. J. M., Van Zaalen, Y., Mens, E. J. M., Van Balkom, H., & Verhoeven, L. (2016a). The concurrent and predictive validity of the Dutch version of the Communicative Development Inventory in children with Down Syndrome for the assessment of expressive vocabulary in verbal and signed modalities. Research in Developmental Disabilities, 56, 99 - 107.

Deckers, S. R. J. M., Van Zaalen, Y., Van Balkom, H., & Verhoeven, L. (submitted). Predictors of receptive and expressive vocabulary development in children with Down Syndrome. Resubmitted with minor revisions to International Journal of Speech Language Pathology.

Deckers, S. R. J. M., Van Zaalen, Y., Stoep, J., Van Balkom, H., & Verhoeven, L. (2016b). Communication performance of children with Down Syndrome: An ICF-CY based multiple case study. Child Language Teaching and Therapy, 32, 293-311. DOI: 0265659016630775.

De Graaf, G. W. (2007). Onderwijs aan leerlingen met Downsyndroom. Deel 1. Down+Up, 79, 46-58.

De Graaf, G. W. (2012). De grote SDS-enquête 2008-2009: Rapportage 2010-2012. Meppel: Stichting Downsyndroom.

De Graaf, G. W., Vis, J. C., Haveman, M., Van Hove, G., de Graaf, E. A., Tijssen, J. G., & Mulder, B. J. (2011). Assessment of prevalence of persons with Down syndrome: A theory-based demographic model. Journal of Applied Research in Intellectual Disabilities, 24(3), 247-262.

De Graaf, G. W., Van Hove, G., & Haveman, M. (2013). More academics in regular schools? The effect of regular versus special school placement on academic skills in Dutch primary school students with Down syndrome. Journal of Intellectual Disability Research, 57(1), 21-38.

Denes, G. (2011). Talking heads: The neuroscience of language. Hove: Psychology Press.

Deniz Can, D., Ginsburg-Block, M., Golinkoff, R. M., & Hirsh-Pasek, K. (2013). A long-term predictive validity study: Can the CDI Short Form be used to predict language and early literacy skills four years later? Journal of Child Language, 40(4), 821-835.

De Ruiter, J. P. (2000). The production of gesture and speech. In D. McNeill. Language and Gesture (pp. 248-311). Cambridge: Cambridge University Press.

de Ruiter, J. P., & de Beer, C. (2013). A critical evaluation of models of gesture and speech production for understanding gesture in aphasia. Aphasiology, 27(9), 1015-1030.

DeVeney, S. L., Hoffman, L., & Cress, C. J. (2012). Communication-based assessment of developmental age for young children with developmental disabilities.Journal of Speech, Language, and Hearing Research,55(3), 695-709.

Dickinson, D. K., McCabe, A., Anastasopoulos, L., Peisner-Feinberg, E. S., & Poe, M. D. (2003). The comprehensive language approach to early literacy: The interrelationships among vocabulary, phonological sensitivity, and print knowledge among preschool-aged children.Journal of Educational Psychology, 95(3), 465.

Dietz, A., Quach, W., Lund, S. K., & McKelvey, M. (2012). AAC assessment and clinical-decision making: The impact of experience. Augmentative and Alternative Communication, 28(3), 148-159.

Dimitrova, N., Özçalışkan, Ş., & Adamson, L. B. (2016). Parents' translations of child gesture facilitate word learning in children with autism, Down syndrome and typical development. Journal of Autism and Developmental Disorders, 46(1), 221-231. doi:10.1007/s10803-015-2566-7

Dodd, B. (1995). Differential diagnosis and treatment of children with speech disorder. Hoboken: John Wiley & Sons.

Dodd, B., & Thompson, L. (2001). Speech disorder in children with Down's syndrome. Journal of Intellectual Disability Research, 45, 308-316.

Dolva, A. S., Coster, W., & Lilja, M. (2004). Functional performance in children with Down syndrome. American journal of occupational therapy,58(6), 621-629.

Dromi, E. (1999). Early lexical development. In M. Barrett (Ed.). The development of language (pp. 99-131). Hove: Psychology Press.

Dugard, P., Todman, J. B., & Staines, H. (2010). Approaching multivariate analysis: A practical introduction. Routledge.

Dunn, L. M., Dunn, L. M., Whetton, C., & Burley, J. NFER-Nelson.(1997). The British Picture Vocabulary Scale Second Edition. UK: NFER-Nelson.

Dykens, E. M. (1995). Measuring behavioral phenotypes: Provocations from the "new genetics". American Journal on Mental Retardation, 99, 522-532.

Dykens, E., Hodapp, R., & Evans, D. (2006). Profiles and development of adaptive behavior in children with Down syndrome.Down Syndrome Research and Practice, 9(3), 45-50.

Eadie, P. A., Fey, M. E., Douglas, J. M., & Parsons, C. L. (2002). Profiles of grammatical morphology and sentence imitation in children with specific language impairment and Down syndrome. Journal of Speech, Language, and Hearing Research, 45(4), 720-732.

Ebert, S., Lockl, K., Weinert, S., Anders, Y., Kluczniok, K., & Rossbach, H. G. (2013). Internal and external influences on vocabulary development in preschool children. School Effectiveness and School Improvement,24(2), 138-154.

Emmorey, K., Borinstein, H. B., Thompson, R., & Gollan, T. H. (2008). Bimodal bilingualism. Bilingualism: Language and Cognition, 11(01), 43-61.

Eriksson, C. C. (2014). Children's vocabulary development. The role of parental input, vocabulary composition and early communicative skills. Stockholm: Department of Special Education Stockholm University.

Fallon, K. A., Light, J. C., & Paige, T. K. (2001). Enhancing vocabulary selection for preschoolers who require augmentative and alternative communication (AAC). American Journal of Speech-Language Pathology, 10, 81-94. doi:10.1044/1058-0360(2001/010)

Farrant, B., & Zubrick, S. (2011). Early vocabulary development: the importance of joint attention and parent-child book reading. First Language, 32, 343–364.

Feldman, H. M., Dale, P. S., Campbell, T. F., Colborn, D. K., Kurs-Lasky, M., Rockette, H. E., & Paradise, J. L. (2005). Concurrent and predictive validity of parent reports of child language at ages 2 and 3 years. Child Development, 76(4), 856-868.

Fenson, L., Dale, P. S., Reznick, J. S., Thal, D., Bates, E., Hartung, J. P., Pethick, S., & Reilly, J. S. (1993). The MacArthur Communicative Development Inventories: User's guide and technical manual. San Diego: Singular Publishing Group.

Fenson, L., Marchman, V. A., Thal, D., Dale, P. S., Bates, E. & Reznick, J. S. (2007). The MacArthur-Bates Communicative Development Inventories user's guide and technical manual, 2nd ed. Baltimore, MD: Brookes.

Fernald, A., & Mazzie, C. (1991). Prosody and focus in speech to infants and adults. Developmental Psychology, 27(2), 209.

Fidler, D. J. (2005). The emerging Down syndrome behavioral phenotype in early childhood: Implications for practice. Infants & Young Children, 18(2), 86-103.

Fidler, D. J., Most, D. E., & Philofsky, A. D. (2008). The Down syndrome behavioural phenotype: Taking a development approach. Down Syndrome Research and Practice, 12, 37–44.

Fidler, D. J., & Nadel, L. (2007). Education and children with Down syndrome: neuroscience, development, and intervention. Mental Retardation and Developmental Disabilities Research Reviews, 13(3), 262-271.

Finke, E. H., & Quinn, E. (2012). Perceptions of communication style and influences on intervention practices for young children with AAC needs. Augmentative and Alternative Communication, 28(2), 117-126.

Foley, K. R., Jacoby, P., Girdler, S., Bourke, J., Pikora, T., Lennox, N., & Leonard, H. (2013). Functioning and post-school transition outcomes for young people with Down syndrome. Child: Care, Health and Development, 39(6), 789-800.

Fowler, A. E. (1990). Language abilities in children with Down syndrome: Evidence for a specific syntactic delay. In D. Cicchetti and M. Beeghly (Eds.). Children with Down syndrome: A developmental perspective (pp. 302-328). Cambridge: Cambridge University Press.

Fröhlich, A. D., & Haupt, U. (2004). Leitfaden zur Förderdiagnostik mit schwerstbehinderten Kindern. Dortmund: Borgmann Publishing.

Galeote, M., Checa, E., Sánchez-Palacios, C., Sebastián, E., & Soto, P. (2016). Adaptation of the MacArthur-Bates Communicative Development Inventories for Spanish children with Down Syndrome: Validity and reliability data for vocabulary. American Journal of Speech-Language Pathology, 25(3), 371-380.

Galeote, M., Sebastián, E., Checa, E., Rey, R., & Soto, P. (2011). The development of vocabulary in Spanish children with Down syndrome: Comprehension, production, and gestures. Journal of Intellectual and Developmental Disability, 36(3), 184-196.

Galeote, M., Soto, P., Checa, E., Gómez, A., & Lamela, E. (2008). The acquisition of productive vocabulary in Spanish children with Down syndrome. Journal of Intellectual and Developmental Disability 33(4), 292-302.

Gathercole, S. E., Pickering, S. J., Hall, M., & Peaker, S. M. (2001). Dissociable lexical and phonological influences on serial recognition and serial recall. Quarterly Journal of Experimental Psychology: Human Experimental Psychology, 54A, 1-30.

Gathercole, S. E., Willis, C. S., Emslie, H., & Baddeley, A. D. (1992). Phonological memory and vocabulary development during the early school years: a longitudinal study. Developmental Psychology, 28(5), 887.

Gentner, D. (1982). Why nouns are learned before verbs: Linguistic relativity versus natural partitioning. In S. Kuczaj (Ed.), Language development: Language, cognition, and culture (pp. 301–334). Hillsdale, NJ: Erlbaum.

Gentner, D., & Boroditsky, L. (2001). Individuation, relativity and early word learning. In M. Bowerman & S. Levinson (Eds.), Language acquisition and conceptual development. New York: Cambridge University Press.

Gillham, B. (1990). First words in normal and Down syndrome children: a comparison of content and word-form categories. Child Language Teaching and Therapy, 6(1), 25-32.

Goldberg, E. (2009). The new executive brain: Frontal lobes in a complex world. Oxford: Oxford University Press.

Golden, C. J. (2004). The Adult Luria-Nebraska Neuropsychological Battery. In G. Goldstein, S. R. Beers, & M. Hersen (Eds.), Comprehensive handbook of psychological assessment, Vol. 1: Intellectual and neuropsychological assessment (pp. 133-146). Hoboken: John Wiley & Sons.

Goldin-Meadow, S. (2007). Pointing sets the stage for learning language—and creating language. Child Development, 78(3), 741-745.

Guralnick, M. J., Connor, R. T., & Johnson, L. C. (2011). The peer social networks of young children with Down syndrome in classroom programmes. Journal of Applied Research in Intellectual Disabilities, 24(4), 310-321.

Hagoort, P. (2007). The memory, unification, and control (MUC) model of language. In T. Sakamoto (ed.). Communicating skills of intention (pp. 259-291). Tokyo: Hituzi Syobo.

Hauser, M. D., Chomsky, N., & Fitch, W. T. (2002). The faculty of language: what is it, who has it, and how did it evolve? Science, 298(5598), 1569-1579.

Hegde, M. N . (2003). Clinical research in communicative disorders: Principles and strategies (3rd ed.). Austin, TX: PRO-ED, Inc.

Hesketh, L. J., & Chapman, R. S. (1998). Verb use by individuals with Down syndrome. American Journal on Mental Retardation, 103(3), 288-304. doi: 10.1352/0895-8017(1998)103<0288:VUBIWD>2.0.CO;2

Hick, R. F., Botting, N., & Conti-Ramsden, G. (2005). Short-term memory and vocabulary development in children with Down syndrome and children with specific language impairment. Developmental Medicine & Child Neurology, 47(08), 532-538.

Hill, K., Kovacs, T., & Shin, S. (2015). Critical issues using brain-computer interfaces for augmentative and alternative communication. Archives of Physical Medicine and Rehabilitation, 96(3), S8–S15. doi:10.1016/j.apmr.2014.01.034

Hills, T. T., Maouene, M., Maouene, J., Sheya, A., & Smith, L. (2009). Longitudinal analysis of early semantic networks preferential attachment or preferential acquisition?. Psychological Science, 20(6), 729-739.

Hodapp, R. M. (1997). Direct and indirect behavioral effects of different genetic disorders of mental retardation. American Journal on Mental Retardation, 102(1), 67-79.

Hoff, E. (2003). The specificity of environmental influence: socioeconomic status affects early vocabulary development via maternal speech. Child Development, 74(5), 1368-1378.

Hoff, E., & Naigles, L. (2002). How children use input to acquire a lexicon. Child Development, 73(2), 418-433.

Hohenberger, A., & Peltzer-Karpf, A. (2009). Language learning from the perspective of nonlinear dynamic systems. Linguistics, 47(2), 481-511.

Huizinga, M., & Smidts, D. P. (2012). BRIEF Vragenlijst executieve functies voor 5-tot 18-jarigen. Amsterdam: Hogrefe Uitgevers.

Hula, W. D., McNeil, M. R., & Sung, J. E. (2007). Is there an impairment of language-specific attentional processing in aphasia? Brain and Language, 103(1), 240-241.

Inui, N., Yamanishi, M., & Tada, S. (1995). Simple reaction times and timing of serial reactions of adolescents with mental retardation, autism, and Down syndrome. Perceptual and Motor Skills, 81, 739-745.

Iverson, J. M. (2010). Developing language in a developing body: the relationship between motor development and language development. Journal of Child Language, 37(02), 229-261.

Iverson, J. M., Longobardi, E., & Caselli, M. C. (2003). Relationship between gestures and words in children with Down's syndrome and typically developing children in the early stages of communicative development. International Journal of Language & Communication Disorders, 38(2), 179-197.

Iverson, J. M., Longobardi, E., Spampinato, K., & Cristina Caselli, M. (2006). Gesture and speech in maternal input to children with Down's syndrome. International Journal of Language & Communication Disorders, 41, 235-251. doi:10.1080/13682820500312151

Jackendoff, R. (2002). Foundations of language: Brain, meaning, grammar, evolution. Oxford: Oxford University Press.

Jackson-Maldonado, D., Thal, D., Marchman, V., Bates, E., & Gutiérrez-Clellen, V. (1993). Early lexical development in Spanish-speaking infants and toddlers.Journal of Child Language,20(03), 523-549.

Jarrold, C., & Baddeley, A. (2001). Short-term memory in Down syndrome: Applying the working memory model. Down Syndrome Research and Practice, 7(1), 17-23.

Kaczmarek, L. A. (2002). Assessment of social-communicative competence: An interdisciplinary model. In H. Goldstein, L. A. Kaczmarek and K. M. English (Eds.). Promoting social communication: Children with developmental disabilities from birth to adolescence (pp. 55-115). Baltimore: Paul H. Brookes Publishing Company.

Kaiser, A. P., Hester, P. P., & McDuffie, A. S. (2001). Supporting communication in young children with developmental disabilities. Mental Retardation and Developmental Disabilities Research Reviews, 7(2), 143-150.

Karmiloff-Smith, A., Al-Janabi, T., D'Souza, H., Groet, J., Massand, E., Mok, K., ... & Tybulewicz, V. (2016). The importance of understanding individual differences in Down syndrome. F1000Research, 5.

Kay-Raining Bird, E., Chapman, R. S., & Schwartz, S. E. (2004). Fast mapping of words and story recall by individuals with Down syndrome. Journal of Speech, Language, and Hearing Research, 47(6), 1286-1300.

Kay-Raining Bird, E., & Cleave, P. (2016). Mothers' talk to children with Down Syndrome, language impairment, or typical development about familiar and unfamiliar nouns and verbs. Journal of Child Language, 43, 1072-1102.

Kay-Raining Bird, E., Cleave, P. L., & McConnell, L. (2000). Reading and phonological awareness in children with Down Syndrome: A longitudinal study. American Journal of Speech-Language Pathology, 9(4), 319-330.

Kay-Raining Bird, E., Gaskell, A., Babineau, M. D., & Macdonald, S. (2000). Novel word acquisition in children with down syndrome: Does modality make a difference? Journal of Communication Disorders,33(3), 241-266.

Kent, R. D., & Vorperian, H. K. (2013). Speech impairment in Down syndrome: A review. Journal of Speech, Language, and Hearing Research, 56, 178-210. doi:10.1044/1092-4388(2012/12-0148)

Kita, S., & Özyürek, A. (2003). What does cross-linguistic variation in semantic coordination of speech and gesture reveal?: Evidence for an interface representation of spatial thinking and speaking. Journal of Memory and language, 48(1), 16-32.

Klee, T., Pearce, K., & Carson, D. K. (2000). Improving the positive predictive value of screening for developmental language disorder. Journal of Speech, Language, and Hearing Research, 43(4), 821-833.

Korkman, M., Kirk, U., & Kemp, S. (2007). NEPSY-II: A developmental neuropsychological assessment. San Antonio: The Psychological Corporation.

Koster, C., Been, P. H., Krikhaar, E. M., Zwarts, F., Diepstra, H. D., & Van Leeuwen, T. H. (2005). Differences at 17 months productive language patterns in infants at familial risk for Dyslexia and typically developing infants. Journal of Speech, Language, and Hearing Research, 48(2), 426-438.

Kouri, T. (1989). How manual sign acquisition relates to the development of spoken language: A case study. Language, Speech, and Hearing Services in Schools, 20(1), 50-62.

Kovacs, T., & Hill, K. (2015). A tutorial on reliability testing in AAC language sample transcription and analysis. Augmentative and Alternative Communication, 31(2), 148-158.

Kronk, R. A., Ogonowski, J. A., Rice, C. N., & Feldman, H. M. (2005). Reliability in assigning ICF codes to children with special health care needs using a developmentally structured interview. Disability and Rehabilitation, 27(17), 977-983.

Kühn, B., & Langner, M. (2012). Experimentum mundi, or a concrete utopia for a language centre in higher education: Building blocks for successful language learning. In B. Kühn and M. Cavana, Perspectives from the European Language Portfolio (pp. 161 – 186). Abingdon: Routledge.

Kumin, L. (1994). Intelligibility of speech in children with Down syndrome in natural settings: Parents' perspective. Perceptual and Motor Skills, 78(1), 307-313.

Kumin, L. (2003). Early communication skills for children with Down Syndrome. Bethesda, MD: Woodbine House.

Kumin, L. (2006). Speech intelligibility and childhood verbal apraxia in children with Down syndrome. Down Syndrome Research and Practice, 10, 10-22.

Kumin, L. (2008). Helping children with Down Syndrome communicate better. Bethesda, MD: Woodbine House.

Kurland, J. (2011). The role that attention plays in language processing. SIG 2 Perspectives on Neurophysiology and Neurogenic Speech and Language Disorders, 21(2), 47-54.

Laws, G. (1998). The use of nonword repetition as a test of phonological memory in children with Down syndrome. Journal of Child Psychology and Psychiatry, 39(8), 1119-1130.

Lederberg, A., & Beal-Alvarez, J. S. (2010). Expressing meaning: From prelinguistic communication to building vocabulary. In M. Marschark and P. Spencer, The Oxford handbook of deaf studies, language, and education (pp. 258-275). Oxford: Oxford University Press.

Lee, D. Y. W. (2001). Defining core vocabulary and tracking its distribution across spoken and written genres. Evidence of a gradience of variation from the British national corpus. Journal of English Linguistics, 29, 250–278. doi:10.1177/00754240122005369

Lee, J. (2011a). Size matters: Early vocabulary as a predictor of language and literacy competence. Applied Psycholinguistics, 32(1), 69-92.

Lee, A. M. (2011b). Using the ICF-CY to organise characteristics of children's functioning. Disability and Rehabilitation, 33(7), 605-616.

Leppänen, P. H., Lyytinen, H., Choudhury, N., & Benasich, A. A. (2004). Neuroimaging measures in the study of Specific Language Impairment. In L. Verhoeven & H. van Balkom (eds) Classification of developmental language disorders (pp. 99-136). Mahwah, NJ: Lawrence Erlbaum.

Levelt, W. J. M. (1989). Speaking: From intention to articulation. Cambridge, MA: MIT Press.

Levelt, W. J. M. (1992). Psycholinguistics: An overview. In W. Bright (Ed.), International Encyclopedia of Linguistics (Second Edition/Vol. 3, pp. 290-294). Oxford University Press.

Levelt, W. J. M. (1993). Speaking: From intention to articulation. Cambridge, MA: MIT press.

Levelt, W. J. M. (1994). What can a theory of normal speaking contribute to AAC? In ISAAC '94 Conference Book and Proceedings (pp.18-20). Toronto: International Society for Augmentative and Alternative Communication.

Levelt, W. J. M. (1999). Producing spoken language: a blueprint of the speaker. In P. Hagoort & C. M. Brown (Eds.) The Neurocognition of Language (pp. 94-122). Oxford: Oxford University Press.

Levelt, W. J. M. (2004). Speech, gesture and the origins of language. European Review, 12, 543-549.

Light, J. (1989). Toward a definition of communicative competence for individuals using augmentative and alternative communication systems. Augmentative and Alternative Communication, 5(2), 137-144.

Light, J., & McNaughton, D. (2015). Designing AAC research and intervention to improve outcomes for individuals with complex communication needs. Augmentative and Alternative Communication, 31(2), 85-96.

Lum, J. A., Conti-Ramsden, G., Page, D., & Ullman, M. T. (2012). Working, declarative and procedural memory in specific language impairment. Cortex, 48(9), 1138-1154.

Maassen, B., Van Haaften, L., & Diepeveen, S. (2014) Computer Articulation Instrument (CAI). Amsterdam: Boom Test Uitgevers.

MacCathren, R. (2000). Teacher-implemented prelinguistic communication intervention. Focus on Autism and Other Developmental Disabilities, 15(1), 21–29.

Mashal, N., Andric, M., & Small, S. L. (2012). Motor and non-motor language representations in the brain. In M. Faust (Ed.), The Handbook of the Neuropsychology of Language (pp. 276-293). Hoboken: John Wiley & Sons..

McCathren, R. B., Yoder, P. J., & Warren, S. F. (1999). The relationship between prelinguistic vocalization and later expressive vocabulary in young children with developmental delay. Journal of Speech, Language, and Hearing Research, 42(4), 915-924.

McDaniel, J., & Yoder, P. J. (2016). Pursuing precision Speech-Language therapy services for children with Down syndrome. In Seminars in Speech and Language, 37, 239-251.

Marvin, C., Beukelman, D., & Bilyeu, D. (1994). Vocabulary-use patterns in preschool children: Effects of context and time sampling. Augmentative and Alternative Communication, 10, 224-236. doi:10.1080/07434619412331276930

Martin, G. E., Klusek, J., Estigarribia, B., & Roberts, J. E. (2009). Language characteristics of individuals with Down syndrome. Topics in Language Disorders, 29(2), 112.

McCauley, R. J. & Fey, M. E. (2006). Treatment of language disorders in children. Baltimore, MD: Brookes.

McLeod, S. (2006). An holistic view of a child with unintelligible speech: Insights from the ICF and ICF-CY. International Journal of Speech-Language Pathology, 8, 293–315.

McLeod, S. (2007). The international guide to speech acquisition. Clifton Park, NY: Thomson Delmar Learning.

McLeod, S. & McCormack, J. (2007). Application of the ICF and ICF-children and youth in children with speech impairment. Seminars in Speech and Language, 28, 254–64.

McLeod, S. & Threats, T. T. (2008). The ICF-CY and children with communication disabilities. International Journal of Speech-Language Pathology, 10, 92–109.

Mein, R., & O'Connor, N. (1960). A study of the oral vocabularies of severely subnormal patients. Journal of Intellectual Disability Research, 4, 130 – 143. doi:10.1111/j.1365-2788.1960.tb00761.x

Melby-Lervåg, M., Lervåg, A., Lyster, S. A. H., Klem, M., Hagtvet, B., & Hulme, C. (2012). Nonword-repetition ability does not appear to be a causal influence on children's vocabulary development. Psychological Science, 23(10) 1092–1098.

Mervis, C. B., & Becerra, A. M. (2003). Lexical development and intervention.Speech and Language Intervention in Down Syndrome, 63-85.

Metsala, J. L., & Walley, A. C. (1998). Spoken vocabulary growth and the segmental restructuring of lexical representations: Precursors to phonemic awareness and early reading ability. In J. L. Metsala & L. C. Ehri (Eds). Word recognition in beginning literacy (pp. 89-120). Mahwah, NJ: Lawrence Erlbaum Associates Publishers.

Mildner, V. (2008). The cognitive neuroscience of human communication. New York, NY: Lawrence Erlbaum.

Millar, D. C., Light, J. C., & Schlosser, R. W. (2006). The impact of augmentative and alternative communication intervention on the speech production of individuals with developmental disabilities: a research review. Journal of Speech, Language, and Hearing Research, 49(2), 248-264.

Miller, J. (1989). Assessing language production in children. Baltimore: University Park Press.

Miller, J. F. (1999). Profiles of language development in children with Down syndrome. In J. F. Miller, M. Leddy & L.A. Leavitt (Eds.), Improving the Communication of People with Down Syndrome. Baltimore: Paul H. Brookes.

Miller, J.F., Leddy, M. & Leavitt, L.A. (1999). Improving the Communication of People with Down Syndrome. Baltimore, MD: Paul H. Brookes.

Miller, J. F., Miolo, G., Sedey, A., & Murray-Branch, J. (1993). The emergence of multiword combinations in children with Down syndrome. Poster presented at the Symposium for Research in Child Language Disorders, Madison, WI.

Miller, J. F., Sedey, A. L., & Miolo, G. (1995). Validity of parent report measures of vocabulary development for children with Down syndrome.Journal of Speech, Language, and Hearing Research,38(5), 1037-1044.

Moore, D., Oates, J., Hobson, R., & Goodwin, J. (2002). Cognitive and social factors in the development of infants with Down syndrome. Down Syndrome Research and Practice, 8(2), 43-52.

Morris, J. K., & Alberman, E. (2009). Trends in Down's syndrome live births and antenatal diagnoses in England and Wales from 1989 to 2008: analysis of data from the National Down Syndrome Cytogenetic Register.BMJ,339, b3794.

Mosse, E. K., & Jarrold, C. (2011). Evidence for preserved novel word learning in Down syndrome suggests multiple routes to vocabulary acquisition. Journal of Speech, Language, and Hearing Research, 54(4), 1137-1152.

Muluk, N. B., Bayoğlu, B., & Anlar, B. (2014). Language development and affecting factors in 3-to 6-year-old children. European Archives of Oto-Rhino-Laryngology, 271(5), 871-878.

Murray, L. L., Holland, A. L., & Beeson, P. M. (1997). Auditory processing in individuals with mild aphasia: a study of resource allocation. Journal of Speech, Language, and Hearing Research, 40(4), 792-808.

Næss, K. A. B., Lyster, S. A. H., Hulme, C., & Melby-Lervåg, M. (2011). Language and verbal short-term memory skills in children with Down syndrome: A meta-analytic review. Research in Developmental Disabilities, 32(6), 2225-2234.

Næss, K. A. B., Melby-Lervåg, A., Lyster, S. A. H., & Hulme, C. (2015). Longitudinal relationships between language and verbal short-term memory skills in children with Down syndrome. Journal of Experimental Child Psychology, 135, 43-55.

Næss, K. A. B., Nygaard, E., Ostad, J., Dolva, A. S., & Lyster, S. A. H. (2016). The profile of social functioning in children with Down syndrome. Disability and Rehabilitation, 1-12.

Nash, H., & Heath, J. (2011). The role of vocabulary, working memory and inference making ability in reading comprehension in Down syndrome. Research in Developmental Disabilities, 32(5), 1782-1791.

Nash, H. M., & Snowling, M. J. (2008). Semantic and phonological fluency in children with Down syndrome: atypical organization of language or less efficient retrieval strategies? Cognitive Neuropsychology, 25(5), 690-703.

Neil, N., & Jones, E. A. (2016). Communication intervention for individuals with Down syndrome: Systematic review and meta-analysis. Developmental Neurorehabilitation, 1-12.

Nijland, L. (2003). Developmental apraxia of speech: Deficits in phonetic planning and motor programming. Nijmegen: Proefschrift Katholieke Universiteit Nijmegen

Oliver, B., & Buckley, S. (1994). The language development of children with Down syndrome: first words to two-word phrases. Down Syndrome Research and Practice, 2(2), 71-75.

Özçalişkan, Ş., Adamson, L. B., Dimitrova, N., Bailey, J., & Schmuck, L. (2016). Baby sign but not spontaneous gesture predicts later vocabulary in children with Down Syndrome. Journal of Child Language, 43(4), 948-63. doi:10.1017/S030500091500029X

Özçalışkan, Ş., & Goldin-Meadow, S. (2011). Is there an iconic gesture spurt at 26 months. In G. Stam and M. Ishino (Eds.). Integrating gestures: The interdisciplinary nature of gesture (pp. 163 – 174). Amsterdam: John Benjamins.

Pan, B., Rowe, M. L., Spier, E., & Tamis-Lemonda, C. (2004). Measuring productive vocabulary of toddlers in low-income families: Concurrent and predictive validity of three sources of data. Journal of Child Language, 31(3), 587-608.

Patterson, T., Rapsey, C. M., & Glue, P. (2013). Systematic review of cognitive development across childhood in Down syndrome: implications for treatment interventions. Journal of Intellectual Disability Research, 57(4), 306-318.

Pepper, J. & Weitzman, E. (2004). It takes two to talk: A practical guide for parents of children with language delays. Toronto: Hanen Centre.

Perez-Pereira, M., & Resches, M. (2011). Concurrent and predictive validity of the Galician CDI. Journal of Child Language, 38(1), 121-140.

Pierce, P., Summer, G., & O'deKirk, M. (2005). The Bridge: A portfolio rating scale of preschool literacy development. Retrieved online on September, 15, 2012.

Pinker, S., & Jackendoff, R. (2005). The faculty of language: what's special about it?. Cognition, 95(2), 201-236.

Polišenská, K., & Kapalková, S. (2014). Language profiles in children with Down Syndrome and children with Language Impairment: implications for early intervention. Research in Developmental Disabilities, 35(2), 373-382.

Power, E., Anderson, A., & Togher, L. (2011). Applying the WHO ICF framework to communication assessment and goal setting in Huntington's Disease: A case discussion. Journal of Communication Disorders, 44, 261–75.

Price, J. R., & Kent, R. D. (2008). Increasing speech intelligibility in Down syndrome and fragile X syndrome. In J .E. Roberts, R. S. Chapman, and S. F. Warren (Eds.), Communication and language intervention series: Speech and language development and intervention in Down syndrome and fragile X syndrome (pp. 219-231). Baltimore: Paul H. Brookes.

Proctor, L. A., & Zangari, C. (2009). Language assessment for students who use AAC. In G. Soto & C. Zangari (Eds.), Practically speaking: Language, literacy, and academic development for students with AAC needs (pp. 47–70). Baltimore, MD: Paul H. Brookes.

Posner, M. I., & Rothbart, M. K. (2007). Research on attention networks as a model for the integration of psychological science. Annu. Rev. Psychol., 58, 1-23.

Posner, M. I., & Fan, J. (2008). Attention as an organ system. In J. R. Pomerantz (Ed.), Topics in integrative neuroscience (pp. 31-61). Cambridge: Cambridge University Press.

Raghavendra, P., Bornman, J., Granlund, M., & Björck-Åkesson, E. (2007). The World Health Organization's International Classification of Functioning, Disability and Health: implications for clinical and research practice in the field of augmentative and alternative communication. Augmentative and Alternative Communication, 23(4), 349-361.

Raghavendra, P., Olsson, C., Sampson, J., Mcinerney, R., & Connell, T. (2012). School participation and social networks of children with complex communication needs, physical disabilities, and typically developing peers. Augmentative and Alternative Communication, 28(1), 33-43.

Ratner, N. B., & Silverman, S. (2000). Parental perceptions of children's communicative development at stuttering onset. Journal of Speech, Language, and Hearing Research, 43(5), 1252-1263.

Reichling, A. (1967). Het woord. Een studie omtrent de grondslag van taal en taalgebruik. Zwolle: Willink.

Renvall, K., Nickels, L., & Davidson, B. (2013). Functionally relevant items in the treatment of aphasia (part I): Challenges for current practice. Aphasiology, 27, 636–650. doi:10.1080/02687038.2013.786804

Riccio, C. A., Sullivan, J. R., & Cohen, M. J. (2010). Neuropsychological assessment and intervention for childhood and adolescent disorders. John Wiley & Sons.

Rice, M. L., Warren, S. F., & Betz, S. K. (2005). Language symptoms of developmental language disorders: An overview of autism, Down syndrome, fragile X, specific language impairment, and Williams syndrome. Applied Psycholinguistics, 26(01), 7-27.

Roberts, R. E., Attkisson, C. C., & Rosenblatt, A. (1998). Prevalence of psychopathology among children and adolescents. American Journal of Psychiatry, 155(6), 715-725.

Roberts, J., Long, S. H., Malkin, C., Barnes, E., Skinner, M., Hennon, E. A., & Anderson, K. (2005). A comparison of phonological skills of boys with fragile X syndrome and Down syndrome.Journal of Speech, Language, and Hearing Research, 48(5), 980-995.

Roberts, J. E., Price, J., & Malkin, C. (2007). Language and communication development in Down syndrome.Mental Retardation and Developmental Disabilities Research Reviews,13(1), 26-35.

Robillard, M., Mayer-Crittenden, C., Minor-Corriveau, M., & Bélanger, R. (2014). Monolingual and bilingual children with and without primary language impairment: Core vocabulary comparison. Augmentative and Alternative Communication, 30, 267–278. doi:10.3109/07434618.2014.921240

Rogers, S. L., Gordon, C. Y., Schanzenbacher, K. E., & Case-Smith, J. (2001). Common diagnoses in pediatric occupational therapy. In J. Case-Smith: Occupational therapy for children (pp. 136–187). St. Louis, MO: Mosby.

Roizen, N. J. (2002). Medical care and monitoring for the adolescent with Down syndrome. Adolescent Medicine, 13, 345–58.

Romski, M. A., & Sevcik, R. A. (2003). Augmented input: Enhancing communication development. In J. C. Light JC, D. R. Beukelman and J. Reichle (Eds.), Communicative competence for individuals who use AAC: From research to effective practice (pp. 147-162). Baltimore, MD: Paul H. Brookes.

Rondal, J. A. (2001). Language in mental retardation: Individual and syndromic differences, and neurogenetic variation. Swiss Journal of Psychology, 60(3), 161-178.

Rondal, J. A., & Edwards, S. (1997). Language in mental retardation. London: Whurr.

Rosin, M. M., Swift, E., Bless, D., & Vetter, D. K. (1988). Communication profiles of adolescents with Down syndrome. Communication Disorders Quarterly, 12(1), 49-64.

Rowland, C. & Fried-Oken, M. (2010). Communication matrix. Journal of Pediatric Rehabilitation Medicine, 3, 319–29.

Rowland, C., Fried-Oken, M., Steiner, S. A., Lollar, D., Phelps, R., Simeonsson, R. J., & Granlund, M. (2012). Developing the ICF-CY for AAC profile and code set for children who rely on AAC. Augmentative and Alternative Communication, 28(1), 21-32.

Sacks, B., & Buckley, S. (2003). Motor development for individuals with Down syndrome–An overview. Cumbria, UK: Down Syndrome Education International.

Salley, B., Panneton, R. K., & Colombo, J. (2013). Separable attentional predictors of language outcome. Infancy, 18(4), 462-489.

Sarimski, K. (2002). Analysis of intentional communication in severely handicapped children with Cornelia-de-Lange syndrome. Journal of Communication Disorders, 35(6), 483-500.

Sato, M., Cavé, C., Ménard, L., & Brasseur, A. (2010). Auditory-tactile speech perception in congenitally blind and sighted adults. Neuropsychologia, 48(12), 3683-3686.

Schiariti, V., Klassen, A. F., Cieza, A., Sauve, K., O'Donnell, M., Armstrong, R., & Mâsse, L. C. (2014). Comparing contents of outcome measures in cerebral palsy using the International Classification of Functioning (ICF-CY): a systematic review. European Journal of Paediatric Neurology, 18(1), 1-12.

Schlichting, L. & Lutje Spelberg, H. (2010a). Schlichting test voor taalbegrip [Schlichting test for language comprehension. Houten, Netherlands: Bohn Stafleu van Loghum.

Schlichting, L. & Lutje Spelberg, H. (2010b). Schlichting test voor taalproductie II [Schlichting test for language production II]. Houten, Netherlands: Bohn Stafleu van Loghum.

Schlosser, R. W., & Raghavendra, P. (2004). Evidence-based practice in augmentative and alternative communication. Augmentative and Alternative Communication, 20(1), 1-21.

Schlosser, R. W., & Wendt, O. (2008). Effects of augmentative and alternative communication intervention on speech production in children with autism: A systematic review. American Journal of Speech-Language Pathology, 17(3), 212-230.

Schmid, H. (2007). Entrenchment, salience and basic levels. In D. Geeraerts and H. Cuyckens (Eds.), The Oxford handbook of cognitive linguistics (pp. 117-138). Oxford: Oxford University Press.

Scholte, E., van Duijn, G., Dijkxhoorn, Y., Noens, I., & van Berckelaer-Onnes, I. (2008) Handleiding Vineland Screener 0–6 jaar [Users manual: Vineland Screener 0–6 years of age]. PITS: Leiden.

Segalowitz, S. J., & Lane, K. C. (2000). Lexical access of function versus content words. Brain and Language, 75, 376-389. doi: 10.1006/brln.2000.2361

Sevcik, R. A. & Romski, M. A. (2002). The role of language comprehension in establishing early augmented conversations. In J. Reichle, D. R. Beukelman, and J. C. Light (Eds.). Exemplary practices for beginning communicators (pp. 453-474). Baltimore, MD: Paul H. Brookes; 2002.

Sheng, L., & McGregor, K. K. (2010). Lexical–semantic organization in children with specific language impairment. Journal of Speech, Language, and Hearing Research, 53(1), 146-159.

Sherman, S. L., Allen, E. G., Bean, L. H., & Freeman, S. B. (2007). Epidemiology of Down syndrome. Mental Retardation and Developmental Disabilities Research Reviews, 13, 221-227. doi:10.1002/mrdd.20157

Silverman, W. (2007). Down syndrome: Cognitive phenotype. Mental Retardation and Developmental Disabilities Research Reviews, 13, 228–36.

Simeonsson, R. J. (2003). Classification of communication disabilities in children: Contribution of the ICF. International Journal of Audiology, 42, S2–8.

Simeonsson, R. J. (2014). Disability determination: Contribution of the ICF-CY. Lecture presented to the Social Security Income Children's Disability Program Committee Meeting, Washington, DC.

Simeonsson, R. J., Lee, A., Granlund, M., & Björck-Åkesson, E. (2010). Developmental and health assessment in rehabilitation with the international classification of functioning, disability and health for children and youth. In: Mpofu E and Oakland T (eds) Rehabilitation and health assessment: Applying ICF guidelines (pp. 27 – 46). New York: Springer.

Simeonsson, R. J., Scarborough, A. A., & Hebbeler, K. M. (2006). ICF and ICD codes provide a standard language of disability in young children. Journal of Clinical Epidemiology, 59(4), 365-373.

Smith, M., & Grove, N. (1999). The bimodal situation of children learning language using manual and graphic signs. In F. Loncke, J. Clibbens, H. Arvidson, & L. Loyd (Eds.), Augmentative and Alternative Communication: New Directions in Research and Practice (pp. 8 – 30). Londol: Whurr.

Smith, B. L., & Oller, D. K. (1981). A comparative study of pre-meaningful vocalizations produced by normally developing and Down's syndrome infants. Journal of Speech and Hearing Disorders,46(1), 46-51.

Spender, Q., Dennis, J., Stein, A., Cave, D., Percy, E., & Reilly, S. (1995). Impaired oral-motor function with Down's syndrome: A study of three twin pairs. European Journal of Disorders of Communication, 30, 77–87.

Sperber, D. & Wilson, D. (1995). Relevance. Oxford: Blackwell.

Sperber, D. & Wilson, D. (1986). Relevance: communication and cognition. Oxford: Blackwell.

Spitzer, R. L., Gibbon, M. E., Skodol, A. E., Williams, J. B., & First, M. B. (2002). DSM-IV-TR Casebook. Arlington, VA: American Psychiatric.

Stefanini, S., Caselli, M. C., & Volterra, V. (2007). Spoken and gestural production in a naming task by young children with Down syndrome. Brain and Language, 101(3), 208-221.

Stoel-Gammon,C. (1997). Phonological development in Down syndrome.Mental Retardation and Developmental Disabilities, 3, 300-306.

Stoel-Gammon, C. (2001). Down syndrome phonology: Developmental patterns and intervention strategies. Down Syndrome Research and Practice, 7(3), 93-100.

Stuart, S., Beukelman, D., & King, J. (1997). Vocabulary use during extended conversations by two cohorts of older adults. Augmentative and Alternative Communication,13, 40 - 47. doi:10.1080/07434619712331277828

Sullivan, J. R., Winter, S. M., Sass, D. A., & Svenkerud, N. (2014). Assessing growth in young children: A comparison of raw, age-equivalent, and standard scores using the Peabody Picture Vocabulary Test. Journal of Research in Childhood Education, 28(2), 277-291.

Taylor, C. L., Christensen, D., Lawrence, D., Mitrou, F., & Zubrick, S. R. (2013). Risk factors for children's receptive vocabulary development from four to eight years in the Longitudinal Study of Australian Children. PLOS one, 8(9), e73046.

Te Kaat, D. J. A. (2013). Expressieve woordenschat bij Downsyndroom [Expressive vocabulary in Down syndrome]. Nederlands Tijdschrift voor Logopedie, 85, 6-10.

The R Foundation (n.d.). The R project for statistical computing. Retrieved November 29, 2016 from https://www.r-project.org/.

Threats, T. T., & Worrall, L. (2004). Classifying communication disability using the ICF. Advances in Speech Language Pathology, 6(1), 53-62.

Tomasello, M. (2003). Constructing a language: A usage-based theory of language acquisition. Cambridge: Harvard University Press.

Tomasello, M. (2008). Origins of Human Communication. Cambridge, MA: MIT Press.

Tomasello, M. & Slobin D. I. (2005). Beyond nature-nurture: Essays in honor of Elizabeth Bates. Mahwah: Lawrence Erlbaum Associates.

Trembath, D., Balandin, S., & Togher, L. (2007). Vocabulary selection for Australian children who use augmentative and alternative communication. Journal of Intellectual and Developmental Disability, 32, 291–301. doi:10.1080/13668250701689298

Ulvund, S. E., & Smith, L. (1996). The predictive validity of nonverbal communicative skills in infants with perinatal hazards. Infant Behavior and Development, 19(4), 441-449.

Üstün, T. B., Chatterji, S., Bickenbach, J., Kostanjsek, N., & Schneider, M. (2003). The International Classification of Functioning, Disability and Health: a new tool for understanding disability and health. Disability and Rehabilitation, 25(11-12), 565-571.

Van Balkom, H. (2009). Communicatie op eigen wijze [Communication in your own way]. Antwerpen: Acco.

Van Balkom, H., Deckers, S. R. J. M., & Stoep, J. (2016/2017). Assessment of communicative competence in children with Severe Developmental Disorders. In E. Segers and P. van den Broek (Eds.). Developmental Perspectives in Written Language and Literacy. Amsterdam: John Benjamins.

Van Balkom, H., & Welle Donker-Gimbrère, M. (2004). Kiezen voor communicatie. Een handboek over de communicatie van mensen met een motorische of meervoudige beperking. Nijkerk: HB Uitgevers.

Van Balkom, H. & Verhoeven, L. (2010). Literacy learning in users of AAC: A neurocognitive perspective. Augmentative and Alternative Communication, 26, 149–57.

Van Balkom, H., Verhoeven, L., Van Weerdenburg, M., & Stoep, J. (2010). Effects of parent-based video home training in children with developmental language delay. Child Language Teaching and Therapy, 26, 221-237. doi:10.1177/0265659009349978

Van Bysterveldt, A. K., Gillon, G., & Foster-Cohen, S. (2010). Integrated speech and phonological awareness intervention for pre-school children with Down syndrome. International Journal of Language & Communication Disorders, 45(3), 320-335.

Vandereet, J. (2010). Longitudinal study of the communication development in young children with intellectual disabilities. Leuven: Proefschrift Katholieke Universiteit Leuven.

Vandereet, J., Maes, B., Lembrechts, D., & Zink, I. (2011). Expressive vocabulary acquisition in children with intellectual disability: Speech or manual signs?. Journal of Intellectual and Developmental Disability, 36(2), 91-104.

Van Duijn, G., Dijkxhoorn, Y., Noens, I., Scholte, E., & van Berckelaer-Onnes, I. (2009). Vineland Screener 0–12 years research version (NL). Constructing a screening instrument to assess adaptive behaviour.International Journal of Methods in Psychiatric Research, 18(2), 110-117.

Van Duijn, G., Dijkxhoorn, Y., Scholte, E. M., & Berckelaer-Onnes, V. (2010). The development of adaptive skills in young people with Down syndrome. Journal of Intellectual Disability Research, 54(11), 943-954.

Van der Meulen, S., Slofstra-Bremer, C., & Lutje-Spelberg, H. (2013). Communicatieve Intentie Onderzoek. Houten: Bohn Stafleu van Loghum.

Van Tatenhove, G. (2009). The Pixon language and learning activity notebook. In Van Tatenhove, G. (Ed.), Pixon project kit. Wooster, OH: Prentke-Romich Company.

Van Tilborg, A., & Deckers, S.R.J.M. (2016). Vocabulary selection in AAC: Application of core vocabulary in atypical populations. ASHA SIG 12: Perspectives on Augmentative and Alternative Communication, 1, 125-138.

Van Tilborg, A., Segers, E., Van Balkom, H., & Verhoeven, L. (2014). Predictors of early literacy skills in children with intellectual disabilities: A clinical perspective. Research in Developmental Disabilities, 35(7), 1674-1685.

Van Zaalen-op 't Hof, Y. (2009). Differential diagnostic characteristics between cluttering and stuttering. Journal of Fluency Disorders, 34, 137-154.

Van Zaalen, Y. & Reichel, I. (2015). Cluttering: Current views on its nature, diagnosis, and treatment. Bloomington: iUniverse.

Verhoeven, L., & Vermeer, A. (2001). Taaltoets Alle Kinderen (TAK). Arnhem: CITO.

Vicari, S., Caselli, M. C., & Tonucci, F. (2000). Asynchrony of lexical and morphosyntactic development in children with Down syndrome.Neuropsychologia, 38(5), 634-644.

Viera, A. J. & Garrett, J. M. (2005). Understanding interobserver agreement: The Kappa statistic. Family Medicine, 37, 360–63.

Viholainen, H., Ahonen, T., Cantell, M., Lyytinen, P., & Lyytinen, H. (2002). Development of early motor skills and language in children at risk for familial dyslexia. Developmental Medicine & Child Neurology, 44(11), 761-769.

Visootsak, J., Hess, B., Bakeman, R., & Adamson, L. B. (2013). Effect of congenital heart defects on language development in toddlers with Down syndrome. Journal of Intellectual Disability Research, 57(9), 887-892.

Vloedgraven, J., Keuning, J., & Verhoeven, L. (2009). Screeningsinstrument beginnende geletterdheid [Screening tool emergent literacy]. Arnhem, Netherlands: Cito.

Wadman, R., Durkin, K., & Conti-Ramsden, G. (2011). Close relationships in adolescents with and without a history of specific language impairment. Language, Speech, and Hearing Services in Schools, 42(1), 41-51.

Washington, K. N. (2007). Using the ICF within speech-language pathology: Application to developmental language impairment. Advances in Speech-Language Pathology, 9, 242–55.

Watt, N., Wetherby, A., & Shumway, S. (2006). Prelinguistic predictors of language outcome at 3 years of age. Journal of Speech, Language, and Hearing Research, 49(6), 1224-1237.

Weighton, C., & Dodd, S. (2011). Treloar vocabulary: A project to meet the AAC needs of teenagers and young adults. Communication Matters, 29, 7 – 9.

Wechsler, D. (2002) Wechsler Preschool and Primary Scale of Intelligence-Third edition. San Antonio: The Psychological Corporation.

Wesche, M., & Paribakht, T. S. (1996). Assessing Second Language Vocabulary Knowledge: Depth Versus Breadth. Canadian Modern Language Review, 53(1), 13-40.

Westby, C. (2007). Application of the ICF in children with language impairments. Seminars in Speech and Language, 28, 265–72.

Wetherby, A. M., & Prizant, B. M. (1993). Profiling communication and symbolic abilities in young children. Communication Disorders Quarterly, 15(1), 23-32.

Wetherby, A. M., Allen, L., Cleary, J., Kublin, K., & Goldstein, H. (2002). Validity and reliability of the communication and symbolic behavior scales developmental profile with very young children. Journal of Speech, Language, and Hearing Research, 45(6), 1202-1218.

Wetherby, A. M., Schuler, A.L., & Prizant, B. M. (1997). Enhancing language and communication development: Theoretical foundations. In D. J. Cohen & F. R. Volkmar (eds). Handbook of autism and pervasive developmental disorders (pp. 513–538). New York: John Wiley and Sons.

Windsor, J., & Fristoe, M. (1991). Key word signing: perceived and acoustic differences between signed and spoken narratives. Journal of Speech, Language, and Hearing Research, 34(2), 260-268.

Wishart, J. G. (1993). The development of learning difficulties in children with Down's syndrome. Journal of Intellectual Disability Research, 37(4), 389-403.

Witkowski, D., & Baker, B. (2012). Addressing the content vocabulary with core: Theory and practice for nonliterate or emerging literate students. Perspectives on Augmentative and Alternative Communication, 21, 74–81. doi:10.1044/aac21.3.74

World Health Organization. (2001). International classification of functioning, disability and health: ICF. Geneva: World Health Organization.

World Health Organization. (2007). International classification of functioning, disability, and health: Children and youth version: ICF-CY. Geneva: World Health Organization.

Wright, C. A., Kaiser, A. P., Reikowsky, D. I., & Roberts, M. Y. (2013). Effects of a naturalistic sign intervention on expressive language of toddlers with Down syndrome. Journal of Speech, Language, and Hearing Research,56(3), 994-1008.

Wullink, M., Van Schrojenstein Lantman-de Valk, H. M. J., Dinant, G. J., & Metsemakers, J. F. M. (2007). Prevalence of people with intellectual disability in the Netherlands. Journal of Intellectual Disability Research, 51(7), 511-519.

Yaruss, J. S. (2007). Application of the ICF in fluency disorders. Seminars in Speech and Language, 28, 312–22.

Ylvisaker, M., & Feeney, T. (2008). Helping children without making them helpless: Facilitating development of executive self-regulation in children and adolescents. In V. Anderson, R. Jacobs, and P. J. Anderson. Executive functions and the frontal lobes: A lifespan perspective (pp. 410-433). New York: Psychology Press.

Yoder, P. J., & Warren, S. F. (2004). Early predictors of language in children with and without Down syndrome. American Journal on Mental Retardation, 109(4), 285-300.

Yoder, P. J., Warren, S. F., Kim, K., & Gazdag, G. E. (1994). Facilitating prelinguistic communication skills in young children with developmental delay II. Systematic replication and extension. Journal of Speech, Language, and Hearing Research, 37(4), 841-851.

Yont, K. M., Snow, C. E., & Vernon-Feagans, L. (2003). The role of context in mother–child interactions: An analysis of communicative intents expressed during toy play and book reading with 12-month-olds. Journal of Pragmatics, 35(3), 435-454.

Yorkston, K., Dowden, P., Honsinger, M., Marriner, N., & Smith, K. (1988). A comparison of standard and user vocabulary lists. Augmentative and Alternative Communication,4, 189 - 210. doi:10.1080 /07434618812331274807

Yorkston, K. M., Smith, K., & Beukelman, D. (1990). Extended communication samples of augmented communicators I: A comparison of individualized versus standard single-word vocabularies. Journal of Speech and Hearing Disorders, 55, 217 - 224. doi:10.1044/jshd.5502.217

Zaidman-Zait, A., & Dromi, E. (2007). Analogous and distinctive patterns of prelinguistic communication in toddlers with and without hearing loss. Journal of Speech, Language, and Hearing Research, 50(5), 1166-1180.

Zampini, L., & D'Odorico, L. (2009). Communicative gestures and vocabulary development in 36-month-old children with Down's syndrome. International Journal of Language & Communication Disorders, 44(6), 1063-1073.

Zampini, L., & D'Odorico, L. (2011). Lexical and syntactic development in Italian children with Down's syndrome. International Journal of Language & Communication Disorders, 46(4), 386-396.

Zampini, L., & D'Odorico, L. (2012). Lo sviluppo del vocabolario nei bambini con sindrome di Down: dati per età cronologica e di sviluppo. Psicologia Clinica dello Sviluppo, 16(2), 331-345.

Zampini, L., & D'Odorico, L. (2013). Vocabulary development in children with Down syndrome: longitudinal and cross-sectional data. Journal of Intellectual and Developmental Disability, 38(4), 310-317.

Zampini, L., Fasolo, M., & D'Odorico, L. (2012). Characteristics of maternal input to children with Down syndrome: A comparison with vocabulary size and chronological age-matched groups. First Language, 32, 324-342.

Zenner, E., Speelman, D., & Geeraerts, D. (2014). Core vocabulary, borrowability, and entrenchment: A usage-based onomasiological approach. Diachronica, 31(1), 74–105.

Zink, I., & Lejaegere, M. (2002). N-CDIs: Lijsten voor communicatieve ontwikkeling. Aanpassing en hernormering van de MacArthur CDIs van Fenson et al., 1993. Leuven: Acco.

# Chapter 8

## Nederlandse samenvatting

# Nederlandse samenvatting

Downsyndroom is de meest voorkomende genetische oorzaak van een verstandelijke beperking. De meeste kinderen hebben een lichte tot matige verstandelijke beperking, met een gemiddeld IQ rond de 50 (Chapman & Hesketh, 2000). Beperkingen én vaardigheden die kenmerkend zijn voor een syndroom vormen samen een gedragsfenotype. Het Downsyndroom gedragsfenotype beschrijft relatieve sterktes in het visuele geheugen, het non-verbaal communiceren en het sociale functioneren. Relatief minder goed ontwikkeld zijn het verbale werkgeheugen, het verwerken van verbale en auditieve informatie, de volgehouden aandacht en aspecten van het motorisch functioneren zoals motorische planning, hypotonie en hyperflexibiliteit (Fidler, 2005; Fidler, Most, & Philofsky, 2008). Een vertraagde en verstoorde taal- en spraakontwikkeling is ook een belangrijk onderdeel van het Downsyndroom gedragsfenotype (Roberts, Chapman, & Warren, 2008). De expressieve taalontwikkeling (het uiten) is vaak meer vertraagd dan de receptieve taalontwikkeling (het begrijpen) en wat op grond van de non-verbale intelligentie verwacht zou mogen worden. Gemiddeld genomen produceren kinderen met Downsyndroom hun eerste, gesproken woord rond de leeftijd van één jaar en negen maanden (Stoel-Gammon, 2001), maar sommige kinderen gaan pas rond hun zesde levensjaar praten (Berglund et al., 2001).

Deze vertraging in de receptieve en expressieve taalontwikkeling heeft een duidelijke relatie met vaak aanwezige communicatieproblemen bij kinderen met Downsyndroom. Communicatieve vaardigheden zijn essentieel om te participeren in verschillende (maatschappelijke) contexten, zoals familie, vrienden, school, werk en de buurt. Communicatieve vaardigheden bieden de mogelijkheid om behoeften duidelijk te maken, om informatie te delen, te vragen en te krijgen en bieden de mogelijkheid om sociale relaties aan te gaan (Light, 1989). Het leren van taal is het resultaat van interacties tijdens gezamenlijke activiteiten (Stoel-Gammon, 2001) en is daarmee een sociale en communicatieve aangelegenheid (Akhtar & Tomasello, 1998). De lexicale ontwikkeling, het leren van woorden en de woordenschatontwikkeling, is een fundament van de taalontwikkeling en wordt dan ook vaak als één van de ‘speciale’ componenten van taal aangeduid (Hauser, Chomsky, & Fitch, 2002; Pinker & Jackendoff, 2005). Zonder een bepaald niveau van woordenschatontwikkeling worden de communicatieve mogelijkheden danig beperkt, omdat in onze talige en verbale wereld woorden noodzakelijk zijn voor het overbrengen van een boodschap (Agdam & Sadeghi, 2014). Woordenschatontwikkeling en de communicatieve mogelijkheden van het kind gaan dus hand in hand. Om de ontwikkeling van woordenschat van kinderen

met Downsyndroom goed in kaart te brengen, is het is dan ook van belang om dit in het perspectief van communicatieve mogelijkheden te bezien (Van Balkom, 2009).

Voorgaand onderzoek naar de woordenschatontwikkeling bij kinderen met Downsyndroom laat een aantal beperkingen of problemen in het onderzoeksdesign zien. De belangrijkste zijn: (a) de meeste onderzoeken kijken alleen naar gesproken woorden en andere modaliteiten (vormen waarin gecommuniceerd kan worden) worden buiten beschouwing gelaten, (b) beperkingen in de gebruikte assessment methode om de lexicale ontwikkeling in kaart te brengen, (c) de meeste studies bij kinderen met Downsyndroom hanteren een cross-sectioneel design, waardoor weinig bekend is over de daadwerkelijke ontwikkeling. Bovenstaande punten worden hieronder een voor een toegelicht.

Kinderen met Downsyndroom, zeker op jonge leeftijd, gebruiken vaak veel gesticulaties en gebaren om zichzelf duidelijk te maken (Galeote et al., 2008), zeker als de spraak nog onvoldoende ontwikkeld is. Onderzoeken waarin deze gebaren niet zijn meegenomen als indicatie van de woordenschatontwikkeling geven daarom mogelijk een onvolledig beeld van wat kinderen met Downsyndroom daadwerkelijk laten zien en kunnen.

De meest gebruikte methode in zowel de praktijk als in onderzoek bij jonge kinderen met Downsyndroom, een oudervragenlijst, is de Nederlandse versie van de MacArthur-Bates Communicative Development Inventories (N-CDI). De validiteit van dit meetinstrument om de woordenschat van kinderen met Downsyndroom in kaart te brengen is nog onvoldoende onderzocht, zeker wat betreft het includeren van gebaren. Daarnaast is ook nog weinig bekend over het functionele woordgebruik van kinderen met Downsyndroom. Het gaat hierbij niet alleen om 'welke woorden kennen ze?', zoals vaak alleen gemeten in onderzoek, maar om 'welke woorden gebruiken ze tijdens de communicatie met verschillende personen en in verschillende situaties?'

Een derde beperking van voorgaand onderzoek is dat de meeste woordenschatstudies bij kinderen met Downsyndroom een cross-sectioneel design hebben. Er zijn maar een beperkt aantal longitudinale studies, waardoor er nog weinig inzicht is in mogelijke voorspellers van de lexicale ontwikkeling (Zampini & D'Odorico, 2013). Binnen deze studies is slechts een beperkt aantal mogelijke voorspellende factoren geïncludeerd (Berglund et al., 2001). Vanuit verschillende socio-neurocognitieve studies (bijv. Denes, 2011; Hagoort, 2007) weten we echter dat de taal- en communicatieve ontwikkeling sterk beïnvloed worden door de ontwikkeling van andere ontwikkelingsdomeinen, zoals waarneming, geheugen, aandacht, cognitie, motoriek en mobiliteit én externe factoren in de omgeving van het kind (Van Balkom, 2009). Deze andere ontwikkelingsdomeinen zijn niet

eerder meegenomen in studies naar de lexicale ontwikkeling bij kinderen met Downsyndroom.

Voortvloeiend hieruit zijn voor dit proefschrift de volgende drie onderzoeksvragen opgesteld:

1. Wat is de validiteit van de N-CDI voor het in kaart brengen van de lexicale ontwikkeling van kinderen met Downsyndroom, waarbij zowel gesproken woorden als gebaren zijn meegenomen?
2. Welke woorden gebruiken kinderen met Downsyndroom het meest in hun spontane, dagelijkse communicatie?
3. Wat zijn de voorspellers van de lexicale ontwikkeling van kinderen met Downsyndroom op groepsniveau en hoe kunnen vaardigheden en beperkingen in deze aan taal- en communicatie gerelateerde ontwikkelingsdomeinen in kaart gebracht worden in individuele casus?

## De validiteit van de N-CDI

Het gebruik van gebaren is een belangrijke manier van communiceren van jonge kinderen met Downsyndroom (Özçaliskan, Adamson, Dimitrova, Bailey, & Schmuck, 2016). De eerste woorden die kinderen via een gebaar uiten, zijn vaak ook de eerste woorden die kinderen met Downsyndroom leren uitspreken (Kouri, 1989). Dit laat zien dat er een sterke relatie is tussen het gebruik van gebaren en de gesproken woordontwikkeling bij kinderen met Downsyndroom (Özçaliskan et al., 2016). In Hoofdstuk 2 van dit proefschrift wordt beschreven hoe, in het assessment van de lexicale ontwikkeling van kinderen met Downsyndroom via de N-CDI, rekening kan worden gehouden met deze multi-modale woordontwikkeling. Deze studie laat een goede concurrente en predictieve validiteit zien van een aangepaste versie van de N-CDI, waarin naast gesproken woorden ook de gebaren die kinderen produceren zijn meegenomen. Dit toont dat ouders een belangrijke bron van informatie zijn in het bepalen van de woordenschatontwikkeling. Özçaliskan et al. (2016) vonden al dat als bij het bepalen van de woordenschatgrootte ook gebaren worden meegenomen, de expressieve woordenschatontwikkeling van kinderen met Downsyndroom vergelijkbaar is met die van zich normaal ontwikkelende kinderen gematcht op non-verbale ontwikkelingsleeftijd. Een en ander contrasteert met de vaak beschreven expressieve taalproblemen in het Downsyndroom gedragsfenotype.

## Functioneel woordgebruik

De aangepaste N-CDI, waarin ook gebaren zijn meegenomen, bleek dus een valide meetinstrument. Dit instrument maakt gebruik van de uitgebreide kennis van ouders van de volledige woordenschat, die hun kind gebruikt in verschillende

situaties. Echter, dit biedt geen inzicht in hoe kinderen met Downsyndroom deze woorden daadwerkelijk gebruiken in deze verschillende situaties (Bello, Onofrio, & Caselli, 2014). Een spontane taalanalyse biedt dan, naast de inzet van de N-CDI, een uitgebreider beeld van de lexicale ontwikkeling. In Hoofdstuk 3 van dit proefschrift is daarom gekeken naar de kernwoordenschat van kinderen met Downsyndroom. Onder de kernwoordenschat vallen woorden die gebruikt worden in verschillende situaties, in interactie met verschillende communicatiepartners en ongeacht het onderwerp van gesprek (Van Tilborg & Deckers, 2017). De resultaten in Hoofdstuk 3 laten zien dat de kernwoorden van kinderen met Downsyndroom sterk overeenkomen met de kernwoordenschat van zich normaal ontwikkelende kinderen; deze kernwoorden hebben dezelfde syntactische, semantische en pragmatische functies. Het percentage van inhoudswoorden en dan met name de zelfstandige naamwoorden is wel wat hoger bij kinderen met Downsyndroom. Het lijkt zeer waarschijnlijk dat dit beïnvloed wordt door het gebruik van gebaren, daar gebaren vooral voor inhoudswoorden worden aangeleerd.

## Voorspellers van de lexicale ontwikkeling

Voorspellers van de lexicale ontwikkeling van kinderen met Downsyndroom zijn onderzocht op groepsniveau in een longitudinale studie (Hoofdstuk 4) en op het individuele casusniveau (Hoofdstuk 5). Op groepsniveau zijn voor de ontwikkeling van de expressieve woordenschat verschillende in- en externe factoren geïdentificeerd: (a) het adaptieve niveau van functioneren, ofwel de ontwikkelingsleeftijd van het kind, (b) de receptieve woordenschat, (c) het opleidingsniveau van moeders, (d) het niveau van communicatieve intentie, (e) fonologisch en fonemisch bewustzijn en (f) de groei in het aandachtsvermogen. De receptieve woordenschatontwikkeling werd voornamelijk voorspeld door het adaptieve niveau van functioneren en door de grootte van de receptieve woordenschat eerder in de ontwikkeling. Deze studie, beschreven in Hoofdstuk 4, is één van de eerste longitudinale pogingen om de woordenschatontwikkeling van kinderen met Downsyndroom te voorspellen en is de eerste die een breed scala aan interne en externe factoren heeft meegenomen. De gevonden voorspellers voor lexicale ontwikkeling bij kinderen met Downsyndroom komen sterk overeen met voorspellers die gevonden zijn bij zich normaal ontwikkelende kinderen. Deze studie sluit daarmee aan bij recente onderzoeksresultaten die laten zien dat de woordenschatontwikkeling van kinderen met Downsyndroom voornamelijk vertraagd is in plaats van afwijkend van de normale ontwikkeling (Polisenka, & Kapalková, 2014).

De meeste voorspellers van de lexicale ontwikkeling zijn vastgesteld op basis van groepsstudies. Uit verschillende studies (bijv. Berglund et al., 2001), maar ook uit de resultaten van Hoofdstuk 2, 3 en 4 blijkt een grote individuele variatie in de lexicale ontwikkeling van kinderen met Downsyndroom. Om de stap te maken naar een passende therapie of interventie om de ontwikkeling te stimuleren, is het belangrijk om deze individuele ontwikkeling in kaart te brengen, waarbij het brede scala aan mogelijke interne en externe factoren wordt meegenomen (McDaniel & Yoder, 2016). Hoofdstuk 5 beschrijft dit 'whole person' of 'totale mens' perspectief in een multiple case-studie design, gebaseerd op de ICF-CY (International Classification of Functioning-Child and Youth) functies en anatomische eigenschappen, activiteiten en participatie en externe en persoonlijke factoren. Hoofdstuk 5 toont hoe het assessment eruit zou moeten zien om over al deze functies en factoren, gerelateerd aan de eerder genoemde ontwikkelingsdomeinen (Van Balkom, 2009) én de voorspellers uit Hoofdstuk 4 iets te kunnen zeggen en op welke manier er een individueel profiel van vaardigheden en beperkingen is op te stellen. Het eerder beschreven Downsyndroom gedragsfenotype kan de gevonden individuele verschillen in ICF-CY domeinen in deze multiple case-studie niet verklaren. Het uitvoeren van case-studies, naast groepsstudies, kan van grote waarde zijn, zeker als het gaat over praktijkgericht onderzoek en de complexiteit van de lexicale ontwikkeling bij kinderen met Downsyndroom.

## Implicaties voor de praktijk

De valide, aangepaste versie van de N-CDI biedt logopedisten de mogelijkheid om middels deze oudervragenlijst, op een snelle en (kosten-)efficiënte manier, inzicht te krijgen in de lexicale ontwikkeling. De aangepaste versie van de N-CDI geeft inzicht in welke woorden het kind kan uiten, middels een gesproken woord en/of een gebaar. Duidelijk werd namelijk dat jonge kinderen met Downsyndroom vaak gebruik maken van gebaren in hun communicatie, naast gesproken woorden. Het is van belang om de verschillende modaliteiten mee te nemen in het assessment. Naast het invullen van de aangepaste N-CDI is het belangrijk om inzicht te krijgen in welke woorden kinderen met Downsyndroom frequent gebruiken in hun communicatie. De woordenlijst, zie Hoofdstuk 3, biedt inzicht in woorden die door de dag gebruikt worden, omdat zij ongeacht de context en communicatiepartner worden ingezet. Deze woorden zouden, indien niet of nauwelijks aanwezig in de woordenschat van het kind, een belangrijke plaats kunnen innemen in een woordenschatinterventie.

Dit proefschrift maakt duidelijk dat de lexicale ontwikkeling een communicatief perspectief nodig heeft. Professionals die werken met én ouders van kinderen

met Downsyndroom zouden zich bewust moeten zijn van de verschillende voorspellers en ontwikkelingsdomeinen die de woordenschatontwikkeling van het kind beïnvloeden. In het opzetten van een interventie om de woordenschat of communicatieve ontwikkeling te stimuleren moeten al deze factoren worden meegenomen. Dit vraagt om interprofessionele samenwerking tussen verschillende disciplines, zoals de logopedist, de ergotherapeut, de fysiotherapeut, de leerkracht, de orthopedagoog én de ouders. Gezamenlijk het beeld vormen van het kind en in gezamenlijkheid formuleren van interventiedoelen, waarbij een ieder aan deze doelen kan werken (Buntinx & Bijwaard, 2004). Rowland en collega's (2012) gaven aan dat, om een effectieve interventie vorm te kunnen geven, er een sterke behoefte is aan een model of instrument, onderbouwd met theorie én vanuit de praktijk, waarmee informatie over de verschillende voorspellers van de taal- en communicatieve ontwikkeling geïntegreerd kunnen worden. Rowland en collega's concludeerden dat een dergelijk model of instrument nog niet voorhanden was. Op basis van de ICF-CY wordt in Hoofdstuk 5 getoond hoe een individueel profiel op te stellen is met behulp van toegankelijke assessment methoden. De beschreven methode in Hoofdstuk 5 biedt professionals de handvatten om dit assessment vorm te geven en om individuele vaardigheden en beperkingen van het kind met Downsyndroom in kaart te brengen. Het is essentieel dat professionals verder kijken dan de grenzen van hun eigen discipline en de inzichten van andere professionals integreren in hun eigen klinisch redeneren en handelen.

# Referenties


Agdam, S. J., & Sadeghi, K. (2014). Two formats of word association tasks: A study of depth of word knowledge. *English Language Teaching*, *7*(10), 1-12.

Akhtar, N., & Tomasello, M. (1998). Intersubjectivity in early language learning and use. In S. Braten. *Intersubjective communication and emotion in early ontogeny* (pp. 316-335). Cambridge: Cambridge University Press.

Bello, A., Onofrio, D., & Caselli, M. C. (2014). Nouns and predicates comprehension and production in children with Down syndrome.*Research in Developmental Disabilities*,*35*(4), 761-775.

Berglund, E., Eriksson, M., & Johansson, I. (2001). Parental reports of spoken language skills in children with Down syndrome. *Journal of Speech, Language and Hearing Research, 44,* 179-191.

Buntinx, W. H. E, & Bijwaard, M. (2004). *Professionaliteit in de zorg voor mensen met functiebeperkingen – Kenmerken, rol, voorwaarden*. Utrecht: Vereniging Gehandicaptenzorg Nederland.

Chapman, R. S. & Hesketh, L. J. (2000). Behavioral phenotype of individuals with Down syndrome. *Mental Retardation and Developmental Disabilities Research Reviews, 6*, 84–95.

Denes, G. (2011). *Talking Heads; The neuroscience of language.* Hove, UK: Psychology Press.

Fidler, D. J. (2005). The emerging Down Syndrome behavioral phenotype in early childhood: Implications for practice. *Infants & Young Children, 18*(2), 86–103.

Fidler, D. J., Most, D. E., & Philofsky, A. D. (2008). The Down syndrome behavioural phenotype : Taking a developmental approach. *Down Syndrome Research and Practice*, 37-44.

Galeote, M., Soto, P., Checa, E., Gómez, A., & Lamela, E. (2008). The acquisition of productive vocabulary in Spanish children with Down syndrome.*Journal of Intellectual and Developmental Disability*,*33*(4), 292-302.

Hagoort, P. (2007). The memory, unification, and control (MUC) model of language. In T. Sakamoto (Ed.). *Communicating skills of intention* (pp. 259-291). Tokyo: Hituzi Syobo.

Hauser, M. D., Chomsky, N., & Fitch, W. T. (2002). The faculty of language: what is it, who has it, and how did it evolve? S*cience*, *298*(5598), 1569-1579.

Kouri, T. (1989). How manual sign acquisition relates to the development of spoken language: A case study. *Language, Speech, and Hearing Services in Schools*, *20*(1), 50-62.

Light, J. (1989). Toward a definition of communicative competence for individuals using augmentative and alternative communication systems. *Augmentative and Alternative Communication*, *5*(2), 137-144.

McDaniel, J., & Yoder, P. J. (2016). Pursuing precision Speech-Language therapy services for children with Down syndrome. In *Seminars in Speech and Language, 37*, 239-251.

Özçalişkan, Ş., Adamson, L. B., Dimitrova, N., Bailey, J., & Schmuck, L. (2016). Baby sign but not spontaneous gesture predicts later vocabulary in children with Down Syndrome. *Journal of Child Language, 43*(4), 948-63.

Pinker, S., & Jackendoff, R. (2005). The faculty of language: what's special about it? *Cognition*, *95*(2), 201-236.

Polišenská, K., & Kapalková, S. (2014). Language profiles in children with Down Syndrome and children with Language Impairment: implications for early intervention. *Research in Developmental Disabilities*, *35*(2), 373-382.

Roberts, J. E., Chapman, R. S., & Warren, S. F. (2008). *Communication and language intervention series: Speech and language development and intervention in Down syndrome and fragile X syndrome*. Baltimore: Paul H. Brookes.

Rowland, C., Fried-Oken, M., Steiner, S. A., Lollar, D., Phelps, R., Simeonsson, R. J., & Granlund, M. (2012). Developing the ICF-CY for AAC profile and code set for children who rely on AAC. *Augmentative and Alternative Communication*, *28*(1), 21-32.

Stoel-Gammon, C. (2001). Down syndrome phonology: Developmental patterns and intervention strategies. *Down Syndrome Research and Practice*, *7*(3), 93-100.

Van Balkom, H. (2009). *Communicatie op eigen wijze*. Antwerpen: Acco.

Van Tilborg, A., & Deckers, S. R. J. M. (2016). Vocabulary selection in AAC: Application of core vocabulary in atypical populations. *ASHA SIG 12: Perspectives on Augmentative and Alternative Communication, 1*, 125-138.

Zampini, L., & D'Odorico, L. (2013). Vocabulary development in children with Down syndrome: Longitudinal and cross-sectional data. *Journal of Intellectual and Developmental Disability*, *38*(4), 310-317.

# Chapter 9

## Dankwoord

## Dankwoord

Had je me tien jaar geleden gevraagd wat ik op dit moment aan het doen zou zijn, dan had ik nooit kunnen bedenken dat het antwoord zou zijn: 'De allerlaatste stap zetten voor het afronden van mijn proefschrift.' Tijdens mijn opleiding tot orthopedagoog heb ik altijd gedacht dat mijn hart in de praktijk ligt. Dat is ook absoluut waar, maar mijn hart klopt ook voor het doen van onderzoek. Dan wel onderzoek waarvan resultaten direct de praktijk in kunnen vloeien. Een perfecte combinatie van praktijk en onderzoek heb ik kunnen vinden op de Fontys Paramedische Hogeschool (FPH) in Eindhoven. Daar ben ik erg dankbaar voor. Resultaten van mijn onderzoek kon ik direct doorvertalen naar onderwijs aan studenten, bijscholing aan het werkveld en trainingen aan ouders. Het waren drukke en emotionele jaren, echt niet alles ging over de spreekwoordelijke 'rolletjes', maar de positieve momenten voeren absoluut de boventoon. En nu is het dus klaar!

Klaar! Indien u wilt weten wat het gebaar voor 'klaar' is, kijk dan eens op de achterkant van mijn proefschrift. Ik hoefde niet lang na te denken wat ik wilde voor mijn kaft. Het hele proefschrift gaat over Downsyndroom, over woordenschat en communicatie. De zoektocht naar die ene juiste foto was nog wel een lastige. Ik ben dan ook enorm blij en dankbaar dat ik hiervoor de hulp heb gekregen van de ouders van Pien en Gijs. Zij stonden er direct voor open om, passend bij het verhaal van mijn proefschrift, foto's te laten maken. Heel erg bedankt. Jullie hebben mijn proefschrift, letterlijk, een gezicht gegeven. Ik wil direct ook Kim's Fotografie & Creatie bedanken, voor het maken van de mooie foto's en ook het meedenken hoe deze foto's het mooist zouden uitkomen op mijn kaft.

Natuurlijk had ik dit proefschrift nooit kunnen schrijven zonder alle kinderen met Downsyndroom, hun ouders, leerkrachten, logopedisten, fysiotherapeuten en alle andere betrokkenen bij de kinderen. Heel erg bedankt dat jullie de afgelopen jaren een inkijk hebben gegeven in jullie leven. Ik heb veel van jullie gevraagd, dat besef ik, maar ik hoop dat ik met dit proefschrift en alles wat er uit voort is gevloeid een positieve bijdrage heb kunnen leveren aan de juiste begeleiding van jullie kinderen. Ik wil hier ook graag Stichting Downsyndroom bedanken voor hun betrokkenheid, kritische blik van tijd tot tijd en de mogelijkheid om meerdere artikelen te publiceren in de Down+Up. In het bijzonder bedankt ik graag Gert de Graaf.

Promoveren doe je niet alleen, en gelukkig maar. In de afgelopen jaren heb ik heel wat steun gehad van allemaal lieve collega's, zowel op de Radboud Universiteit als op FPH. Het gaat om zo veel collega's, dat het bijna een boekwerk wordt, als ik iedereen bij naam ga noemen. Alle OLO promovendi, post-docs, docenten en UD's, professoren en natuurlijk iedereen van het secretariaat, heel erg bedankt

voor alle leuke, mooie en kritische momenten in de afgelopen jaren. Bedankt ook dat ik als buiten-promovendus, als ‘de vreemde eend’, me altijd heb thuis gevoeld in jullie midden. Ik ga de afdeling absoluut missen als mijn verdediging is geweest. Tevens een groot woord van dank voor het team Logopedie van FPH, waar ik in 2011 bij kwam aanwaaien. Jullie hebben me opgenomen, altijd op waarde geschat en ook bij jullie heb ik me al snel thuis gevoeld. Datzelfde geldt nu ook voor mijn lieve collega’s van team Interprofessioneel Samenwerken. Bedankt allemaal, ook aan alle (voormalige) MT-leden en directeur(en) en de collega’s van alle andere teams en staf op FPH, dat jullie het in mij zagen en zien zitten en dat ik de ruimte heb gekregen om mezelf te ontwikkelen en mijn promotie-onderzoek tot een goed einde te brengen. Ook wil ik alle studenten bedanken die hebben geholpen, bij dataverzameling, maar ook door het schrijven van hun scripties en daarmee met nieuwe literatuur en inzichten kwamen. De grootste dank gaat uit naar alle studenten van de opleiding Logopedie, in de afgelopen vijf jaar hebben ruim 150 tweede, derde én vierdejaars een rol gespeeld in mijn project.

Promoveren kun je natuurlijk niet zonder promotoren. Mijn dank gaat dan ook absoluut uit naar mijn promotoren Ludo Verhoeven en Hans van Balkom. Ludo, jij had altijd een nuchtere blik, hield het overzicht en kwam met kritische vragen. Jouw overzicht heeft er heel erg aan bijgedragen dat mijn proefschrift gestructureerd werd. Jij hebt altijd de rode draad voor ogen en dat heeft mij erg geholpen. Hans, jouw enorme expertise en het enthousiasme waarmee je dat ventileert werken aanstekelijk. Al vanaf het eerste moment dat ik je ontmoette, was ik ‘verliefd’ op de manier waarop jij met passie over de doelgroep en Ondersteunde Communicatie praat. Je bent een wandelende encyclopedie als het gaat om theorieën met betrekking tot de communicatieve en taalontwikkeling. Ik heb in jou, naast alle praktijkvoorbeelden uit mijn stages en vrijwilligerswerk bij de Wigwam, mijn inspiratie voor dit hele mooie onderwerp gevonden.

Via Hans ben ik betrokken geraakt bij de OC-leerstoel van de Radboud Universiteit. Hier heb ik met heel veel plezier Judith, Mascha en Evelien leren kennen. Met allemaal een andere blik, een ander perspectief, maar allemaal met een enorme gedrevenheid hebben jullie mijn (PhD-)leven verrijkt. Ik denk met heel veel plezier terug aan alle mooie momenten die we hebben meegemaakt, zoals Pittsburgh in 2012 en Lissabon in 2014. Bedankt voor alle steun, goede raad, feedback, motiverende gesprekken, humor en warmte.

Ik had dit allemaal niet kunnen doen, mezelf niet zo kunnen ontwikkelen en ontplooien en nooit dit, in mijn ogen, mooie proefschrift kunnen schrijven zonder de rol van Yvonne, mijn co-promotor. Yvonne, jij hebt mij kritisch leren denken, me zo vaak gewezen op mijn veel te lange zinnen, me er op momenten doorheen gesleept. Maar bovenal, jij hebt mij vanaf moment één op waarde geschat, je

zag het in me zitten, hebt me overal mee naartoe genomen en ik heb zo veel van je geleerd. Behalve het maken van korte zinnen, dat gaat me nog steeds niet zo goed af. We hebben vele mooie momenten gehad, binnen ons RAAK-project, met aanvragen schrijven, tijdens onze communicatieweekenden, het winnen van de Fontys Onderzoeksprijs en onze reis langs verschillende universiteiten in New York en Charlottesville. Te veel om op te noemen en dat lijstje gaan we hopelijk in de volgende jaren absoluut nog uitbreiden.

Arjan en Roy, mijn paranimfen, mijn maatjes, makkers, beste vrienden, Arie en Finch. Op de afdeling OLO werden we wel eens 'de drie musketiers' genoemd. En zo voelt het ook. We hebben elkaar (goed) leren kennen tijdens de Research Master, alweer bijna 8 jaar geleden; het eerste contact met Arjan verliep 'iets' moeizamer dan met Roy, maar gelukkig is alles goed gekomen. We hebben samen de laatste jaren van onze studententijd mogen vieren, erg mooie momenten met Maïzena (d.i. de studievereniging). Er was ook nog iets met een komkommer, maar dat ben ik verder vergeten. Veel avonden samen koken, kletsen, voetbal kijken, bier drinken, koffie drinken in het DE café en dan Kitty en Jo van het werk houden; wat was het dagelijks genieten met jullie. In de woorden van Mascha: "Jullie zijn een cadeau en de momenten met jullie een feestje." We hebben alle drie onze moeilijke en mooie momenten gehad in onze persoonlijke levens in die afgelopen jaren. Jullie waren er altijd voor mij, door dik en dun. Ik weet zeker dat onze sterke band blijft, ook nu ieder inmiddels zijn eigen weg is gegaan.

Mijn laatste woord van dank en van liefde gaat natuurlijk uit naar Kellie. Het was niet altijd makkelijk voor je in die afgelopen jaren en je hebt er wel eens van gebaald: de vele avonden laat thuis van werk en dan ook nog eens op de bank verder werken, de weekenden waarin ik vaak nog achter mijn laptop zat en altijd maar dat gezeur over dingen die niet goed gingen, over "stomme reviewers en tijdschriften." Je hebt me daarin alle ruimte gegeven, me gesteund, me opgepept. Je was er altijd als ik dat nodig had, met een lieve lach, een dikke knuffel, met een lief woord, maar vooral door er gewoon te zijn.

# Chapter 10

## Project and Curriculum Vitae

## Curriculum vitae

Stijn Deckers was born on August 31st , 1987 in Heerlen, The Netherlands. Upon the completion of his high school education at Bernardinus College, he obtained a bachelor degree in Pedagogical Sciences at Radboud University Nijmegen in 2008. Given his interest in the development of children with intellectual and multiple disabilities, he proceeded with a master in Pedagogical Sciences in the focus programme on intellectual, physical and multiple disabilities. In 2009 he received his master degree in Pedagogical Sciences, bene meritum. His interest in research was awakened during the study for his master thesis on feeding problems in toddlers with developmental disabilities, supervised by prof. dr. Jan de Moor and dr. Jan van der Burg. Therefore he chose to proceed with the Research Master of Behavioural Sciences at the Behavioural Science Institute at Radboud University Nijmegen. The research project and thesis for the research master – supervised by prof. dr. Robert Didden – focused on sleep problems in adolescents and adults with a mild to moderate intellectual disability. In 2011 he received his research master degree, bene meritum.

Directly following, Stijn started his PhD project at the School for Allied Health Professions, Fontys University of Applied Sciences, Eindhoven, The Netherlands. Next to his research work, for the past five years he was a lecturer for the departments of Speech Language Pathology and Interprofessional Collaboration at Fontys University. He obtained the University Teaching Qualification (BKO certificate) in 2012. His PhD project – supervised by dr. Yvonne van Zaalen (Fontys University), prof. dr. Ludo Verhoeven and prof. dr. Hans van Balkom (both Radboud University) – was on the lexical development of children with Down syndrome. Additionally, Stijn was involved as senior researcher for the RAAK PRO-project ‘Praten kan ik niet…, maar communiceren wil ik wel!’ [I can’t speak…, but I do want do communicate!’]. Together with dr. Yvonne van Zaalen he wrote a book on Augmentative and Alternative Communication (AAC) and organised the Dutch conference on AAC in 2015. Currently Stijn is still involved in education and research at Fontys University and he is the president of the Dutch and Flanders chapter of the International Society for Augmentative and Alternative Communication (ISAAC-NF).

# List of publications related to PhD

## Journal contributions

### *English articles*

**Deckers, S. R. J. M.**, Van Zaalen, Y., Stoep, J., Van Balkom, H., & Verhoeven, L. (2016). Communication performance of children with Down Syndrome: an ICF-CY based multiple case study. *Journal of Child Language Teaching and Therapy, 32*, 293-311.

**Deckers, S. R. J. M.**, Van Zaalen, Y., Mens, E. J. M., Van Balkom, H., & Verhoeven, L. (2016). The concurrent and predictive validity of the Dutch version of the Communicative Development Inventories in children with Down syndrome for the assessment of expressive vocabulary in verbal and signed modalities. *Research in Developmental Disabilities, 56,* 99-107.

Van Tilborg, A., & **Deckers, S. R. J. M.** (2016). Vocabulary selection in AAC: Application of core vocabulary in atypical populations. *ASHA SIG 12: Perspectives on Augmentative and Alternative Communication, 1, 125-138.*

Van Balkom, H., **Deckers, S. R. J. M.**, & Stoep, J. (2017). Assessment of communicative competence in children with severe developmental disorders. In E. Segers & P. van den Broek (Eds.). *Developmental Perspectives in Written Language and Literacy.* Amsterdam: John Benjamins.

**Deckers, S. R. J. M.**, Van Zaalen, Y., Van Balkom, H. & Verhoeven, L. (2016). Predictors of receptive and expressive vocabulary development in children with Down Syndrome. Reviewed manuscript submitted to *International Journal of Speech-Language Pathology*.

**Deckers, S. R. J. M.,** Van Zaalen, Y., Van Balkom, H., & Verhoeven, L. (2017). Core vocabulary of young children with Down syndrome. Augmentative and Alternative Communication, 33, 77-86.

### *Dutch articles*

**Deckers, S. R. J. M.**, & Van Zaalen, Y. (2012). Een leuke 'tool' om beter te leren praten. *Down & Up, 98,* 29.

**Deckers, S. R. J. M.**, Van der Borgh, L., Brouns, E., & Van Zaalen, Y. (2014). Inzet van communicatiehulpmiddelen leidt tot betere communicatie. *Bosk Magazine*, december 2014.

**Deckers, S. R. J. M.**, & Van Zaalen, Y. (2014). Het in kaart brengen van de ontwikkeling van jonge kinderen met Downsyndroom. *Down&Up*, december 2014.

**Deckers, S. R. J. M.** (2014). ICF-CY en Downsyndroom: is het een bruikbaar middel voor het nemen van beslissingen gericht op interventie in de praktijk? *DigiKomCom, 2*, 22-23.

**Deckers, S. R. J. M.**, Snellens, S., Van Kemenade, L., Lammers, M., Molenbroek, S., Smits, M., Peek, S., & Van Zaalen, Y. (2015). De keuze voor Ondersteunde Communicatie bij kinderen met Downsyndroom: interviews met ouders en logopedisten. *Down+Up, 112,* 33-37.

**Deckers, S. R. J. M.**, Van Kemenade, L., Lammers, M., Molenbroek, S., Smits, M., Snellens,S., Peek, S., & Van Zaalen, Y. (2016). De inzet van Communicatie Ondersteunende Hulpmiddelen bij Downsyndroom: interviews met ouders, logopedisten, leerkrachten en adolescenten/volwassen met Downsyndroom. *Geplaatst in lentenummer 2016 Down+Up.*

Tuinhout, M., & **Deckers, S. R. J. M.** (2016). Kinderen met downsyndroom: de richtlijn. *Podosophia, 24*(1), 22-26.

**Deckers, S. R. J. M.** & Van Zaalen, Y. (2016). Communicatie als fundament van de orthopedagogiek: Ondersteunde Communicatie bij jonge kinderen met Downsyndroom. Orthopedagogiek: Onderzoek en Praktijk, 55 (9-10), 223-232.

## Conference Contributions

### *Presentations and workshops*

**Deckers, S. R. J. M.**, Stoep, J., & Van Zaalen, Y. (2012). Ondersteunde Communicatie bij mensen met een verstandelijke beperking. Lecture presented at conference on Rubinstein-Taybi syndrome, Beekbergen.

**Deckers, S. R. J. M.**, Stoep, J., Van Balkom, H. & Van Zaalen, Y. (2012). RAAK PRO project 'Praten kan ik niet… maar communiceren wil ik wel': een introductie in Ondersteunde Communicatie. Lecture presented at conference on Pitt Hopkins syndrome, Zoetermeer.

**Deckers, S. R. J. M.**, van Zaalen, Y., Peek, M. & Van Balkom, H. (2012). How to implement AAC effectively in SLP. Expert workshop presented at the CPLOL congress, The Hague, Netherlands.

**Deckers, S. R. J. M.**, Van Zaalen, Y., Stoep, J., Van Balkom, H., & Verhoeven, L. (2012). OC (technologie) en het (jonge) kind met Downsyndroom. Interactive workshop presented at conference 'Kracht van Communicatie', ISAAC-nf, Kerkrade.

Stoep, J., **Deckers, S. R. J. M.**, Van Zaalen, Y., Van Balkom, H., & Verhoeven, L. (2012). Profiling communicative competence: A neurosociocognitive reasoning model for AAC assessment. Lecture presented at the ISAAC congress, Pittsburgh, USA.

Van Tilborg, A., Van Wingerden, E., **Deckers, S. R. J. M.**, Van Balkom, H., Segers, E., Verhoeven, L., Stoep, J., & Van Zaalen, Y. (2012). Enhancing (basic) literacy skills in children with intellectual disabilities through AAC. Lecture presented at the ISAAC congress, Pittsburgh, USA.

**Deckers, S. R. J. M.,** & Van Zaalen, Y. (2013). Down Syndrome: One developmental profile? Lecture presented at the Speech Language Therapy Summer School, Patras, Greece.

**Deckers, S. R. J. M.** & Van Zaalen, Y. (2013). ICF-CY en Downsyndroom. Lecture presented at conference NVLF, Nieuwegein.

**Deckers, S. R. J.M**., Van Zaalen, Y., Van Balkom, H., Stoep, J., & Verhoeven, L. (2014). ICF-CY and Down Syndrome: a useful tool for clinical decision making? Lecture presented at the International Society for Augmentative and Alternative Communication (ISAAC) conference, Lisbon, Portugal.

Van Balkom, H., Stoep, J., **Deckers, S. R. J. M**., & Van der Schuit, M. (2014). Profiling communicative competence: a neurosociocognitive reasoning model to guide decision-making in AAC assessment. Lecture presented at the ISAAC conference, Lisbon, Portugal.

**Deckers, S. R. J. M**., Stoep, J., & Van Zaalen, Y. (2014). Insights in vocabulary organization in children with Down Syndrome. Lecture presented at the ISAAC conference, Lisbon, Portugal.

**Deckers, S. R. J. M.** (2014). Levelt's model: from blueprint of the speaker to blueprint of the communicator? Lecture presented during the research symposium, ISAAC conference, Lisbon, Portugal.

**Deckers, S. R. J. M.,** Van Zaalen, Y., & Peek, M. (2014). Interprofessional treatment planning in Down Syndrome: round table discussion. Expert workshop presented at the Speech Language Therapy Summer School, Malta.

Van Zaalen, Y. & **Deckers, S. R. J.M.** (2014). Roadmap to person-to-system matching in Augmentative and Alternative Communication. Lecture presented at the Speech Language Therapy Summer School, Malta.

**Deckers, S. R. J. M.** (2014). Levelt's model: from blueprint of the speaker to blueprint of the (AAC) communicator? Lecture presented during the Pittsburgh AAC Language Seminar Series, Pittsburgh, USA.

**Deckers, S. R. J. M.** & Van der Meulen, J. (2015). Gebruik Communicatieve Intentie Onderzoek bij cliënten met een Ernstige Meervoudige Beperking. Lecture presented for Logopediekring Reinaerde.

Van Zaalen, Y. & **Deckers, S.R.J.M.** (2015). Speech and Fluency development in children with Down Syndrome. Lecture presented at the International Fluency Association conference in Lisbon, Portugal.

**Deckers, S. R. J. M.,** Van Zaalen, Y., & Peek, M. (2015). Interprofessional treatment planning in Down Syndrome: case study in communication problems. Expert workshop presented at the Speech Language Therapy Summer School, Aachen, Germany.

**Deckers, S. R. J. M.** & Baker, B. (2015). Insights from behavioral, psycholinguistic and neurosociocognitive models for the field of AAC. Lecture presented at the Clinical Augmentative and Alternative Communication conference in Charlottesville, Virginia, USA.

Van Balkom, H. & **Deckers, S. R. J. M.** (2015). Profiling Communicative Competence: A neurosociocognitive reasoning model to guide decision-making in AAC Assessment. Lecture presented at the Clinical Augmentative and Alternative Communication conference in Charlottesville, Virginia, USA.

**Deckers, S. R. J. M.** & Van der Meulen, J. (2015). Gebruik Communicatieve Intentie Onderzoek en Communication Matrix (bij VB/MB). Presented at AAC conference: 'Praten kan ik niet…., maar communiceren wil ik wel!', Eindhoven.

**Deckers, S. R. J. M.** (2015). OC en woordenschat(ontwikkeling) bij Downsyndroom. Presented at AAC conference: 'Praten kan ik niet…., maar communiceren wil ik wel!', Eindhoven.

**Deckers, S. R. J. M.** (2016). Downsyndroom en taal- en communicatieontwikkeling: nieuwe inzichten en mogelijkheden. Presented at conference for SO/VSO Daniël de Brouwerschool, Wilp.

**Deckers, S. R. J. M.** & Raadsen, M. (2016). Samenwerken aan een beter doel: interventieplanning in een casus met Downsyndroom tijdens een Interprofessioneel Overleg. Interactive workshops at the Education Festival Interprofessional Collaboration, Hogeschool Utrecht.

**Deckers, S. R. J. M.** & Van Zaalen, Y. (2016). Down syndrome: Speech, language and communication development. Lecture presented at Seton Hall University, New Jersey.

Van Zaalen, Y. & **Deckers, S. R. J. M.** (2016). Interprofessional collaboration & Interprofessional education. Lecture presented at Adelphi University, New York.

**Deckers, S. R. J. M.** & Van Zaalen, Y. (2016). Interprofessional treatment planning in Down Syndrome: case study in communication problems. Interactive workshop presented at Adelphi University, New York.

## *Poster presentations*

**Deckers, S. R. J. M.**, & Van Zaalen, Y. (2011). Gebruik van OC voor stimuleren van communicatieve redzaamheid en beginnende geletterdheid van kinderen met Downsyndroom, Poster presented at ZON MW conference, Focus on Research, Utrecht.

**Deckers, S. R. J. M.**, & Van Zaalen, Y. (2012). Communication Competencies Profile in children with Down Syndrome. Poster presented at the CPLOL conference, The Hague, Netherlands.

**Deckers, S. R. J. M.**, van Zaalen, Y., & Reichel, I. K. (2014). Using the ICF-CY to identify individual strengths and weaknesses in communicative competence in children with Down Syndrome. Touro College Research Day. New York, U.S.A.

Van Zaalen, Y. & **Deckers, S. R. J. M.** (2015). Speech and fluency development in young children with Down Syndrome. Poster presented at the International Fluency Association conference, Lisbon, Portugal.