SPEECH, LANGUAGE AND SLEEP PROBLEMS IN CHILDREN WITH DOWN SYNDROME: A ROLE FOR SPEECH-LANGUAGE PATHOLOGISTS?

by

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We accept this thesis as conforming to the required standard

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ABSTRACT

Language development can be used as a proxy for development. By investigating language development in a clinical model, we may be able to gain deeper insights into the developing brain. In this thesis, I am suggesting a clinical concept for monitoring *speech and language development* with a focus on children with Down syndrome. Children with Down syndrome experience speech and language difficulties as well as sleep problems. The latter may affect their development, including their speech and language abilities. This thesis presents a proposed protocol to investigate interactions. The content seems to be highly relevant because if such an association can be demonstrated, we may be able to improve the speech and language abilities of children with Down syndrome through the early identification and treatment of sleep problems. This would result in improving their wellbeing and quality of life, allowing them to be better integrated into the community and reduce parents' burden.

TABLE OF CONTENTS

LIST OF TABLES

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1

INTRODUCTION

1.1 Language development

Researchers study language development from different perspectives and disciplines, across a variety of university departments. Neurolinguistic research has shown that language development is correlated with changes in brain development and maturation (Sakai 2005; Friederici 2006; Rosselli et al. 2014) in typically developing individuals and individuals with language impairment. Using structural neuroimaging tools (such as magnetic resonance imaging), researchers have found that language, at a macroscopic scale, may be lateralized to both hemispheres of the brain (Kadis et al. 2011). This is likely to persist through childhood (Friederici et al. 2011) and then become more lateralized as age increases (Everts et al. 2009). MRI studies have found that increased lateralization in the left hemisphere is correlated with improved language skills (Hervé et al. 2013), including grammar abilities (Nuñez et al. 2011). Improved language skills in childhood (measured by language testing) are also positively correlated with brain white matter volume and negatively correlated with brain gray matter volume (Rosselli et al. 2014), whereas improved language skills in senescence are positively correlated with brain gray matter volume (Rosselli et al. 2014).

Reviewing the literature, *I have concluded that language development is associated with certain changes in brain development, which I will describe in detail in the next subchapters.*

1.1.1 Speech and language development in typically developing children

Typically developing children follow a pattern when acquiring language. In the phonological (i.e. sound system) domain, young infants are responsive to human voices and to sounds in their environment from birth (Feldman 2005). Young infants can also distinguish between many vowel sounds from their native language shortly after birth (Moon et al. 2013), and this ability becomes more tuned to their native language by their first birthday (Werker and Tees 1984; McQuiston and Kloczko 2011).

Speech development begins first as babbling, where infants produce sounds that are not recognizable as words, during the first year. Babbling serves many functions such as signalling wants (e.g. hunger, thirst) to learning to match their adult input as infants are responding to their environment. Babbling usually ends around the first birthday when the infant starts to produce words. Words become more intelligible to adults as correct articulation of speech sounds is acquired. Of special note, the age at which specific speech sounds are acquired is different for most typically developing children learning English, but the order of acquisition is similar (Sander 1972). They may still have difficulty pronouncing some speech sounds or words when they enter kindergarten. Typically developing children learning English will have mastered the speech sounds and be fully intelligible to adults by age 7-8 years (Feldman 2005). Speech development is assessed in clinical practice using standardized articulation tests (e.g. Goldman-Fristoe Test of Articulation-2) and by sampling speech in single word tests or in conversations to assess articulation of a specific speech sound (ASHA 2014a). In the research setting, other methods are used in addition to standardized tests and sampling natural speech: parent report, elicited production (where children are prompted to answer a question), and repetition tasks to test articulation (Ambridge and Rowland 2013).

Language development in typically developing children learning English occurs in a sequence, most notably in grammar where it emerges in a predictable order (Brown 1973). Production of questions begins when the child produces longer sentences. Questions are normally routinized in the beginning (especially questions with who, what, where) and yes/no questions tend to be made with intonation. The question structure quickly becomes generalized for a variety of asking. Complex sentences with two or more verbs can appear as early as 2 years, but usually take longer to develop. Proper use of pronouns also takes longer to appear due to the complexity of their meanings. Words occurring at higher rates in parental input are acquired earlier because children tend to mirror what they hear (Aguado-Orea and Pine 2015). Other factors affect acquisition as well, such as cognitive development; perceptual salience of words to be acquired; and number of meanings: it is easier to learn a word if it has only one associated meaning, than if it has multiple associated meanings (which introduces ambiguity). By 2-3 years, typically developing children comprehend language very well. Following age 2-3 years, they grow their vocabulary, produce longer sentences and expand their use of gestures to accompany their language output. At age 7-8, typically developing children continue dialogue when interacting with familiar adults (Abe et al. 2013). In their case study report, Abe et al. (2013) found that typically developing children used more dialogue and regulatory skills than storytelling and non-verbal communication skills. Dialogue skills include starting new topics, expressing personal feelings, commenting and maintaining dialogue (Abe et al. 2013); regulatory skills include requesting objects, requesting information and directing attention towards their conversation partner (Abe et al. 2013). This indicates that typically developing children have developed sufficient language abilities to carry conversations with adults. When storytelling at this age, they express the character's emotions, mental states, and thoughts (McKeough 1992). Narratives are also more

complex with regards to grammar and story episodes (Bento and Befi-Lopes 2010). Language development is assessed in clinical practice and in the research setting using standardized language tests (e.g. Preschool Language Scale-5) as well as judgement tasks, where children rate if a sentence is well-formed (Ambridge and Rowland 2013; ASHA 2014b; Schutz 2014). Parent report and observations of language use, including storytelling/narratives, across different situations and environments is also used (ASHA 2014b; Schutz 2014). In addition, in the research setting, comprehension of language is also tested using tasks: e.g. acting out a scene, pointing, preferentially looking at one visual scene or another, preferentially listening to one speaker or another (Ambridge and Rowland 2013).

1.1.2 Speech and language development in children with Down syndrome

Compared to typically developing children, children with Down syndrome have delayed language and speech abilities beyond what is expected for their cognitive delay (Cleland et al. 2010). This delay is well documented in the linguistic and developmental psychology literature. There are deficits in all domains of speech and language, and in both comprehension or "receptive language", and production or "expressive language" (Chapman et al. 1991, 1998; Abbeduto et al. 2007). Specific areas that have been investigated include speech abilities, vocabulary, grammar and writing abilities.

As described above, *speech development* is marked with the onset of babbling. In children with Down syndrome, characteristics and development of babbling are similar to typically developing children (Stoel-Gammon 2001). Despite these similarities, their speech development differs. Speech sound development is delayed (Stoel-Gammon 2001; Roberts et al. 2007) and may not follow the same order as for typically developing children (Kumin et al. 1994). Speech errors

often persist to adolescence and adulthood (Stoel-Gammon 2001). Some individuals with Down syndrome are unintelligible throughout their lifetime (Stoel-Gammon 2001). Speech development in children with Down syndrome is assessed in clinical practice using standardized assessments, parent report, sampling natural speech and analyzing sounds produced, storytelling as well as observations on communication (Schutz 2014). In the research setting, the speech of children with Down syndrome is assessed similarly to clinical practice, with the addition of analyzing sampled speech using a computer acoustics program and computer-assisted analysis of speech (Roberts et al. 2007; Martin et al. 2009; Kent and Vorperian 2013). More time-consuming methods may be additionally used in the research setting, possibly due to increased availability of time compared to clinical practice.

Language development in children with Down syndrome has similarities and differences compared to typically developing children. Roberts et al. (2007) conducted a literature review to characterize the similarities and differences. In the pre-linguistic period, similar to typically developing children, children with Down syndrome use gestures, develop vocalizations and babble. While the pre-linguistic period lasts approximately 12-18 months in typically developing children, it can last for several years or a lifetime in individuals with Down syndrome depending on phenotype. ("Phenotype" is defined as severity of the syndrome.) Beyond the pre-linguistic period, language development in children with Down syndrome progresses at a delayed rate compared to their typically developing peers. Children with Down syndrome are delayed in the production of first words and meaningful speech compared to typically developing peers (Stoel-Gammon 2001). Most notably, it takes longer to acquire semantics (i.e. word meanings) and grammar. Pragmatics (i.e. nonverbal communication abilities) are also delayed but to a lesser extent than semantics and grammar. For example, children with Down syndrome can maintain

topics well during conversation, but provide less detailed information when doing so. Roberts et al. (2007) interpret children with Down syndrome as being ambiguous when talking, but they are able to provide clarification when asked by their communication partner. Abbeduto et al. (2007) also conducted a literature review to characterize the general language development of individuals with Down syndrome. They found that individuals with Down syndrome exhibit strengths in imitation of others, gesture use to support communication, word recognition and sound retrieval. Individuals with Down syndrome also appear to use language as a social tool and have good pragmatic ability. Most weaknesses in language abilities lie in expressive language and sentence ambiguity. The findings of Abbeduto et al.'s (2007) literature review reproduce those of Roberts et al.'s (2007) literature review. Of special note, Abbeduto et al. (2007) also described that the language impairment experienced by individuals with Down syndrome is not fully present at birth and it does change with age: individuals with Down syndrome progress and develop their speech and language, albeit at a slower pace compared to typically developing peers. Language development of children with Down syndrome is assessed in clinical practice and in the research setting using similar methods: parental report, non-standardized clinical activities and use of standardized tests (Schutz 2014). In addition, more elaborate methods such as analysis of conversation abilities (e.g. initiating conversation, interactions, elaboration on topics) and narration may be used for evaluation of vocabulary and grammatical knowledge (Martin et al. 2009).

Reviewing the literature, *I have concluded that children with Down syndrome may be a good clinical model to investigate language development as their language development shows greater variability compared to typically developing children. Thus, information about factors influencing development and abilities is needed with serial measurements.*

In the next subchapter, I will describe the sleep problems experienced by children with Down syndrome as one of the major factors influencing cognitive abilities.

1.2 Sleep

Sleep is a neurological function necessary for somatic, emotional and general well-being (Jan et al. 2008). It is connected to all facets of life and function. Sleep problems transcend cultural boundaries and are noted to occur around the globe in all populations of children (Smaldone et al. 2007). Consequences of sleep disorders range in severity and are associated with medical/physical problems, behavioural concerns and can impact academic ability and success (Smaldone et al. 2007; Mindell and Owens 2009). There are numerous studies that demonstrate associations between sleep and daily function, for example: behaviour problems (Sadeh et al. 2002), working memory (Steenari et al. 2003), and cognitive functioning (Buckhalt et al. 2007). In this subchapter, I will outline the prevalence and sleep problems experienced by typically developing children and by children with neurodevelopmental conditions (with a focus on Down syndrome).

1.2.1 Sleep in typically developing children

Epidemiological studies have found that sleep problems may be prevalent in typically developing populations of children. An estimated 20% to 25% of typically developing children in the USA experience a variety of sleep problems (Smaldone et al. 2007; Krakowiak et al. 2008), with 30% of children experiencing at least transient sleep deprivation (Jan et al. 2008).

On the other hand, sleep problems have a variety of negative consequences and the list of factors with which sleep problems are associated continues to grow. An example is the relationship of sleep problems to obesity (Seicean et al. 2007; Cespedes et al. 2016), with one study reporting

a 41% increase in the odds of childhood obesity for every hour of reduction in sleep duration (Ievers-Landis et al. 2008). Other examples include: association with behaviour problems in toddlers (Goodlin-Jones et al. 2009; Hall et al. 2012) and in school-aged children (Rosen et al. 2004); executive function (Anderson et al. 2009); and sensory processing abnormalities (Vasak et al. 2015). Sleep disordered breathing (a sleep disorder characterized on a spectrum ranging from snoring, to shallow breathing, upper airway obstructions and apnea; Pack 2006) has been associated with increased asthma severity (Ross et al. 2012) and lower cognitive functioning (Gottlieb et al. 2004). In a study of nearly 300 children, sleep disordered breathing was associated with markedly lower levels of health-related quality of life (Rosen et al. 2002).

Reviewing the literature, *I have concluded that sleep problems affect development. To what degree sleep problems are normal and when they affect development remains an open question. Sleep problems are high priorities in parental complaints to physicians (Mindell and Owens 2009). Thus, a clinical model which investigates chronic sleep problems in a high-risk population showing great variability may support finding the answer. My personal take home message is that sleep problems should be screened to inform prevention measures as treatment will improve abilities and most probably quality of life of children.*

1.2.2 Sleep in children with neurodevelopmental conditions and in children with Down syndrome

Children with neurodevelopmental conditions. In contrast to typically developing children, Jan et al. (2008) estimate that 75% to 80% of children with neurodevelopmental conditions experience sleep problems. This is consistent with previous findings, which range from 44% to 86% from samples of children with autism spectrum disorder or other developmental disability (Krakowiak

et al. 2008). The majority of sleep problems in children with neurodevelopmental conditions present as insomnia resulting in chronic sleep deprivation (Malow et al. 2012). Like typically developing children, sleep problems in children with neurodevelopmental conditions are associated with many negative consequences. This has been documented for cognition, daytime behaviour, emotion and mood, executive function, and coordination (Jan et al. 2008; Sivertsen et al. 2012; Esbensen 2016). Sleep problems could additionally impact other aspects of health (e.g. cardiovascular, endocrine, immune, metabolic), which may in turn lead to increased sleep difficulties (Jan et al. 2008). Although highly prevalent, insomnia-associated sleep problems are often undiagnosed and untreated (Malow et al. 2012), as descriptive clinical measures supporting identification of causes do not exist for children with neurodevelopmental and/or mental health conditions (Ipsiroglu et al. 2015).

Children with Down syndrome. Estimates suggest the prevalence of sleep problems in children with Down syndrome range from 31% to 54% (Stores and Stores 2013) to as high as 69% (Esbensen 2016). Similar to other individuals with neurodevelopmental conditions, sleep problems in individuals with Down syndrome may not be thoroughly investigated or their severity may be underestimated (Marcus et al. 1991; Shott 2006; Shott et al. 2006) because individuals with Down syndrome experience a multitude of comorbidities (Charleton et al. 2014). Sleep problems may be regarded as untreatable (Robinson and Richdale 2004; Shott 2006), but if appropriately and properly diagnosed, sleep problems are treatable conditions (Montgomery et al. 2004; Meltzer and Mindell 2007; Stores and Stores 2013; Ipsiroglu et al. 2016). Deleterious effects of sleep problems have been documented in children with Down syndrome (Chen et al. 2013; Esbensen 2016; Lukowski and Milojevich 2017). An in-depth focus on sleep problems in this population may thus be beneficial to improving their overall health and wellbeing.

The main sleep disorder which has been investigated in individuals with Down syndrome is sleep disordered breathing. Individuals with Down syndrome are greater risk than the average person to develop this disorder, including airway obstruction (Shott 2006; Lal et al. 2015), due to physical characteristics of the syndrome (stature, neck and anatomical characteristics of upper airways). All children with Down syndrome are recommended to have a sleep study by the age of 3 to 4 years to assess this comorbidity (Committee on Genetics 2001; Shott et al. 2006). If diagnosed, sleep disordered breathing is treated with nasal sprays, by surgical removal of the tonsils and/or adenoids, or with non-invasive ventilation during sleep (Shott 2006; Lal et al. 2015).

Sleep problems in this population may also be multifactorial in aetiology (Stores and Stores 2013). Parental reports have described observation-based sleep problems that can be broadly classified into three main categories based on reported day- and night-time behaviours (Stores and Stores 2013):

(1) Excessive daytime sleepiness (EDS). EDS has been reported extensively by parents in survey studies (Carter et al. 2009; Breslin et al. 2011; Ashworth et al. 2013). Children with Down syndrome have also been found to take more naps during the day than typically developing peers (Stores 1993).

(2) Insomnia (difficulties falling asleep and staying asleep). Carter et al. (2009) found that compared to the typically developing children, children with Down syndrome have difficulties falling asleep: the children in their study displayed defiant and oppositional behaviour at bedtime and consequently took longer to fall asleep. The authors also found that once asleep, the children frequently have difficulties staying asleep: the children in their study had nighttime awakenings and difficulties going back to sleep after waking up (Carter et al. 2009).

(3) Parasomnias. Parasomnias are "undesirable behavioural, physiological or experiential events that accompany sleep" (Markov et al. 2006); e.g. night terrors, teeth grinding, sleep walking, sleep talking, but also nightmares. Parasomnias have also been reported by parents of children with Down syndrome (reviewed in Stores and Stores 2013). In Carter et al.'s (2009) study, the most common parasomnias were teeth grinding and sleep talking, with very few individuals experiencing night terrors or sleepwalking.

Investigations of other sleep problems are also underway. One example is rhythmic movement disorder, a neurological disorder characterized by repetitive movements such as head banging and body rocking before or during sleep (Khan et al. 2008). The prevalence of rhythmic movement disorder in children with Down syndrome has not yet been estimated and is being investigated as of 2015 (Health Research Authority of the UK National Health Service). A second example is epilepsy, a neurological disorder characterized by recurrent seizures (Fisher 2015). Its prevalence in Down syndrome may range from 1% to 13% (Stafstrom and Konkol 1994; Arya et al. 2011) and epilepsy may affect sleep quality (Jain and Kothare 2015). How it affects the sleep and development in individuals with Down syndrome remains an open question as studies are missing. Circadian rhythms may be robust in infants and young children with Down syndrome (Fernandez et al. 2017), but further research is needed to determine to what degree this is similar in older individuals. In addition, there are sleep problems, such as Restless Legs Syndrome (RLS), which have not been investigated until recently. From my knowledge, the first presentation in poster form came from the Sleep/Wake-Behaviours Clinic and Research Lab of BC Children's Hospital, which I am integrated in (Chan et al. 2015). RLS is a neurological disorder characterized by an "urge-to-move" and discomfort (up to pain) in the feet, legs, hands, arms and/or other body parts which

worsens during periods of rest and towards the nighttime (Ipsiroglu et al. 2016). RLS is a frequent cause of insomnia in adults (Ipsiroglu et al. 2016), and it affects 2% to 4% of children and adolescents (Picchietti et al. 2007; Yilmaz et al. 2011). The prevalence of RLS in children with Down syndrome has not yet been determined. In our clinical study of RLS in children with Down syndrome, we found that 100% (16/16) children experienced symptoms of RLS and 94% (15/16) experienced insomnia (Chan et al. 2015). RLS seems to play a significant role. Thus, given the overall increased risk for sleep problems, the American Academy of Pediatrics suggests that all children with Down syndrome be assessed regularly for sleep problems at doctor health maintenance visits (Committee on Genetics 2001).

Reviewing the literature, *I have concluded that children with Down syndrome experience sleep problems caused by a variety of factors. The prevalence and early appearance of sleep problems may affect their development. Further research is needed to determine the magnitude of effect that sleep problems may have. A beginning step is described below with the results of the Down Syndrome Needs Survey.*

1.3 Results of the Down Syndrome Needs Survey

The Down Syndrome Needs Survey was conducted in 2015 to gain a better understanding of the current health statuses of individuals with Down syndrome in Canada. The survey was developed at the Sleep/Wake-Behaviours Clinic and Research Lab of BC Children's Hospital in collaboration with the Down Syndrome Research Foundation (DSRF) (Burnaby, Canada), the Down Syndrome Clinic and Research Centre at the Kennedy Krieger Institute (Baltimore, USA) and healthcare professionals including paediatricians, medical specialists and speech-language pathologists (UBC C&W #: H14-02305) (Beyzaei et al. CMAJ, submitted, 2017.). The survey comprised five

sections: (i) demographics; (ii) diagnoses medications, and supplements; (iii) development (including speech and language development); (iv) sleep/wake-behaviours; (v) feedback and testimonials regarding healthcare service delivery. I assisted in the question selection for the 'development' section, and I contributed in developing the survey in the online REDCap format (Research Electronic Data Capture; from the Kids Brain Health Network). I also had the privilege to contribute to five abstracts and posters (Chan et al. 2016; Beyzaei et al. 2017a, 2017b; Tse et al. 2017a, 2017b).

The survey was released in British Columbia from March 15 to April 15, 2015 and was advertised online by the Down Syndrome Research Foundation. For the last two weeks of data collection, it was opened to families in Alberta, Ontario and other provinces. 349/364 responses with Canadian postal codes were analyzed. As the survey comprised multiple sections, the total number of respondents per question may vary. This was accounted for in the following presentation of results. For analyses, the data is separated by age (<6, 6-12, 13-18, >18 years).

The majority of individuals with Down syndrome in our sample experience difficulties with language (76%). The results of the survey are presented in **Tables 1 to 3**.

Table 1: Percentage of individuals who experience difficulties with language; n=268.

	Age Separation n (%)[1]				
Experiences difficulties	<6 years	6-12 years	13-18 years	>18 years	**Average**
With either expressive and/or receptive language	59(81)	62(82)	28(82)	55(65)	**204(76)**
With only expressive language	29(40)	26(34)	8(24)	13(15)	**76(28)**
With only receptive language	4(5)	3(4)	1(3)	11(13)	**19(7)**
With both expressive and receptive language	26(36)	33(43)	19(56)	31(36)	**109(41)**

[1] Percentages reflect the correct n-value for each cell.

Table 1 presents the language difficulties experienced by individuals with Down syndrome.

Table 2: Parent/caregiver-reported average sentence length when speaking and/or signing; n=196.

	Age Separation n (%)[1]				
Average length of sentence	<6 years	6-12 years	13-18 years	>18 years	**Average**
No words (speech or sign) yet	8(12)	2(3)	2(7)	2(5)	**14(7)**
1 word	31(48)	5(8)	0(0)	5(11)	**41(21)**
2 words	10(15)	8(13)	1(4)	1(2)	**20(10)**
3 words	10(15)	18(30)	8(30)	4(9)	**40(20)**
4 words	3(5)	9(15)	4(15)	9(20)	**25(13)**
5 words or more	3(5)	18(30)	12(44)	21(48)	**54(27)**
Unsure of sentence length	0(0)	0(0)	0(0)	2(5)	**2(1)**

[1] Percentages reflect the correct n-value for each cell. Percentages may not add to 100% in each column due to rounding.

Table 2 presents the average sentence length when speaking/signing (equivalent to a parental estimate of the "mean length of utterance" used in linguistic research).

Table 3: Symptoms of sleep disorders[1] experienced by individuals with expressive and/or receptive language difficulties as reported by parents/caregivers; n=197[2].

Symptoms	Age Separation n (%) <6 years	6-12 years	13-18 years	>18 years	Average
Sleep disordered breathing:					
1 symptom[3]	51(89)	53(90)	24(86)	44(83)	**172(87)**
2+ symptoms[3]	43(75)	41(69)	19(68)	29(55)	**132(67)**
3+ symptoms[3]	30(53)	32(54)	14(50)	18(34)	**94(48)**
4+ symptoms[3]	14(25)	20(34)	7(25)	7(13)	**48(24)**
Insomnia:					
Wakes up during the night	31(54)	40(68)	15(54)	14(26)	**100(51)**
Problems falling asleep	9(16)	12(20)	13(46)	16(30)	**50(25)**
Parasomnias:					
Teeth Grinding	16(28)	23(39)	11(39)	19(36)	**69(35)**
Sleep Talking/Shouting	9(16)	9(15)	6(21)	13(25)	**37(19)**
Nightmares	6(11)	6(10)	1(4)	2(4)	**15(8)**
Getting up confused	5(9)	5(8)	1(4)	1(2)	**12(6)**
Night terrors	3(5)	3(5)	0(0)	0(0)	**6(3)**
Sleep Walking	0(0)	2(3)	2(7)	1(2)	**5(3)**

[1] Described symptoms by parents/caregivers of individuals with Down syndrome.
[2] N-value is lower as some surveys were incomplete.
[3] Symptoms of sleep disordered breathing include: mouth breathing, snoring, breathing pauses, gasping, bed wetting, and restless sleep.

Table 3 presents the sleep problems experienced by those who have language difficulties.

Of interest was that 37% (97/265) of parents/caregivers reported that the communicative ability of their child, adolescent or adult with Down syndrome was better than other individuals of the same age with Down syndrome, which warrants further investigations. 25% (65/265) reported that they were the same, and 28% (73/265) reported that they were worse. 11% (30/265) were unsure of how their child, adolescent or adult with Down syndrome compared to other individuals of the same age.

To learn further about the speech and language abilities of children with Down syndrome and how to work with the children, I had the opportunity to observe speech therapy sessions with two speech-language pathologists (SLPs) at the DSRF. The three sessions are briefly described below (presented names are pseudonyms):

Session #1: "Cassie" is a 10-year-old girl who has disruptive behaviours, most prominently difficulties taking turns. Her speech is good, but her family would like her to learn cooperative skills as she is entering Grade 2. The first game (mailing letters) focused on taking turns while the second game (baking a cake) focused on sharing. The session ended with singing, a relaxing activity. After the session, her SLP told me that Cassie improved greatly over the course of her sessions and is now allowing others to join her in the activities. Her SLP commented that this supports Cassie's transition in elementary school and her integration with other children.

Session #2: "Taylor" is a 6-year-old boy who has limited language abilities. He goes to speech therapy to practice his speech and to learn language. At a previous session, his SLP introduced an *I want…* sentence strip and he is practicing using it. Taylor says, "I want" followed by pointing to/saying the name of an activity (e.g. a drink, a break, the washroom) on his sentence strip. This enables Taylor to communicate more clearly with his family, teachers and peers because he has a reference structure. Taylor led the activities during the session; we played with the toys in the playroom. After the session, his SLP informed me that because Taylor does not focus well in highly structured settings, she had Taylor practice his speech during playtime to increase participation. Taylor rehearses specific signs and practices saying names of objects and activities using his sentence strip.

Session #3: "Megan" is a 5-year-old girl who has disruptive behaviours. Megan likes to overturn cups and other objects to empty their contents. Her SLP informed me that Megan once overturned a full cup of water and got the entire desk wet. During the session, we played with the toys in the playroom, sang songs and watched YouTube videos on the computer. Megan was full of energy and ran around the room. Her SLP remarked that Megan also does not focus well with rigid structures, so speech practice is embedded into Megan's play. For example, her SLP would ask her to repeat the names of the toys she was playing with. Of special interest, Megan also receives math and reading lessons from a teacher at the DSRF. Her therapy session is followed by math/reading class; the advantage is that her SLP and her teacher can work with her on transitioning from one activity in one location to another activity in a different location. Megan often has difficulties transitioning at school, thus by rehearsing, she is being prepared for these situations.

Observations of the speech therapy sessions and subsequent review of the observations with the SLPs taught me four lessons: **(1) Family involvement.** SLPs work with families to set and achieve goals for clients. Therapy sessions are not meant only for improving speech and language abilities, but also for improving skills such as taking turns, waiting in line, sharing, requesting items, and transitioning from one activity to the next. Therapy priorities depend on the priorities of the family. It is thus important to establish a relationship with parents/caregivers to plan therapy sessions accordingly. **(2) Setting goals.** Goals are important for achievement of and reviewing previous

long term priorities. To increase motivation and participation from the child to reach the set goals, therapy goals are often embedded in play. One example I saw during Session #1 was using a game where "Cassie" was practicing taking turns with others (a major goal for the family) in a game setting. Another example during Session #3 was when the SLP asked "Megan" to name the toys to practice her articulation. **(3) Therapeutic setting.** Therapy can and should be a safe place for clients to learn about the 'real world' and to practice using tools that can be used to enable personal success. For example, during Session #2, the SLP constantly referred "Taylor" to his *I want*... sentence strip and had him practice using it. "Taylor" can then use this tool in his day-to-day life (particularly at school) to communicate his wants and needs. **(4) The role of the SLP.** SLPs work with clients and their families to support their communication needs. Partly, they complement other professionals, services and programs. Aims of therapy can be mutual with the aims of other professionals, and therapy sessions can integrate goals from other programs as appropriate. For example, during Session #3, the DSRF's occupational therapist (OT) often walked in on the session as her office is located next to the SLP offices. "Megan" receives both OT and SLP services, and the therapists worked together during her playtime to have her lift objects (OT aim) but also name the objects (SLP aim). Another example is the SLP working with "Megan's" teacher to practice transitions.

To learn further about the disruptive behaviours of children with Down syndrome, I participated as a research assistant in our clinic's *Sleep & Challenging Behaviours Study* in the summer of 2016 (Campbell et al. 2016). Collaborating with a summer student, I observed the daytime behaviours of children with Down syndrome during the DSRF's summer school program. We observed five children over four weeks of full day classes. All five children experience sleep problems. One personally significant child is briefly described below (presented name is a

pseudonym):

> **"Aiden"** is a 13-year-old boy with an uneven profile who attended two weeks of summer school. He is sensitive and understands the concepts of 'trust' and 'independence'. Aiden reads picture books with ease and his reading teacher constantly looked for higher level material for him to read. Aiden also solves math problems quickly; his math teacher created new math games for him to play (e.g. multiplying and dividing number blocks) and to practice his "money math" skills. Aiden has behaviours perceived as disruptive, such as temper tantrums, interrupting others and running out of the classroom. On one occasion, Aiden ran into the washroom and locked himself in the stall; based on our observations, the comments "I don't trust you" (referring to Aiden) may have triggered this behaviour. He made a mess in the washroom before coming out. Other behaviours displayed included being a "class clown" and self-stimulating by spinning in his chair or playing with markers, which may have been caused by relieving emotional distress.

In general, the summer study (and in particular, my observations of "Aiden") taught me that Down syndrome phenotype varies greatly in severity. For example, "Aiden" demonstrated that he understood the significance of complex ideas and he consistently excelled at all the activities during summer school. This contrasts with other children with a more severe phenotype who are nonverbal and cannot fully participate in activities without help from aides. Our research experience, which occurred in a team setting with Dr. Osman Ipsiroglu (supervisor), Mackenzie Campbell (the summer student) and myself, also taught me how to review behaviours from different perspectives. There are different ways to rank and categorize behaviours; for example, we could use pre-established categories and 'fit' our data to those categories. If we review behaviours from the *child's perspective*, we may gain a different insight into "why" the behaviour occurs. This allows for a complementary concept to highlight recurring themes in the data. For example, in the *Sleep & Challenging Behaviours Study*, we differentiated so-called disruptive behaviours in three themes (Campbell et al. 2016): (1) *challenging behaviours, which have an explanatory model.* This category includes behaviours such as being overly enthusiastic, stubborn, self-conscious or having a temper tantrum. (2) *goofy behaviours, which do not have an explanatory model and come from light-heartedness or silliness.* This category includes behaviours such as

being a "class clown" or distracted with objects. (3) *unspecified behaviours, which do not have an explanatory model.* This category includes behaviours such as self-stimulating, being disengaged or intrusive, and not doing as told. There is no (current) explanation for *why* unspecified behaviours occur. Thus, from the point of view of the child, some behaviours can be explained (1 and 2) while others cannot be explained (3). This concept and these themes highlight the challenges that professional staff may experience in interpretation of behaviours. My research perspective is to make observations reproducible by using video recordings. As I am interested in becoming an SLP, the question is to what degree can SLPs (as facilitators of therapeutic settings) help support children with disruptive behaviours.

***In summary**, my observations at summer school taught me that children with Down syndrome, like any other child, need a safe place to grow and develop. A therapeutic setting, which may be one-to-one (e.g. a therapy session) or in a summer camp, can facilitate this by providing a space for having fun and for exploration of personal traits (e.g. likes/dislikes, aptitude for playing musical instruments, sexuality). To consider different viewpoints, video recordings can be used because they provide a reproducible setting. Observations are discussed and pooled to facilitate a mutual understanding of the children's behaviours.*

1.4 Discussion

Our survey results reproduce key findings from the literature: **(1)** There is a high prevalence of language difficulties among individuals with Down syndrome. This high prevalence is present in high proportions across all age groups, and individuals with Down syndrome tend to have difficulties with both receptive and expressive language (**Table 1**). The findings are consistent with the reviews of Roberts et al. (2007) and Abbeduto et al. (2007), who described not only the

delays but also the difficulties (i.e. factors contributing to communication breakdowns) experienced by individuals with Down syndrome. We note that the incidence of communication breakdowns decreases as language abilities improve (shown by decreasing percentages across the four age groups in **Table 1**).

(2) There are deficiencies in overall communication, as demonstrated by individuals having low parent-reported average sentence lengths (**Table 2**). This finding is consistent with Roberts et al.'s (2007) and Abbeduto et al.'s (2007) reviews, as well as Berglund et al.'s (2001) study which included tracking average sentence lengths in children with Down syndrome aged <6 years. By age 5, typically developing children have an average sentence length of 4 words or more (Brown 1973). Thus, the majority of individuals with Down syndrome aged 6 years to adulthood in the survey should have a parent-reported average sentence length of at least 4 words or 5 or more words, if their language was developing like their typically developing peers. We see that this was not the case (**Table 2**). One possible contributor to these language difficulties is their speech production and voice quality. Kent and Vorperian (2013) reviewed the speech abilities and voice qualities of individuals with Down syndrome and found that voice abnormalities (e.g. pitch differences, increased noise that affect voice quality) are present early in life, although it is not understood if this persists or changes over time. Disfluencies (e.g. stuttering) are also more likely to occur in individuals with Down syndrome, possibly because of phonological processing or motor difficulties and coordination (Kent and Vorperian 2013). Further, 28% of survey respondents also rated the language abilities of their child, adolescent or adult with Down syndrome as worse than other individuals of the same age, indicating that the communication deficits are perceived by parents/caregivers. Parents/caregivers may not always understand what their child, adolescent or adult with Down syndrome is saying due to the presence of speech errors.

Thus, inarticulate pronunciations of a word their child knows may not be included as part of their estimation of average sentence length, leading to a lower estimate. Another contributor to the deficiencies could be their weak grammar skills because weak grammar skills may lead to inabilities to form longer sentences with many words.

(3) There is a high prevalence of sleep problems beginning at an early age (i.e. from <6 years onwards; **Table 3**). This reproduced the results from a study by Bassell et al. (2015), who found that sleep problems, such as bedtime resistance, parasomnias and symptoms of sleep-disordered breathing, begin early from 1 to 3 years. Bassell et al. (2015) found that some sleep problems improve, such as having less (but still significant) bedwetting problems and less difficulties falling asleep; other sleep problems persisted, such as difficulties staying asleep and symptoms of sleep-disordered breathing. This suggests that sleep affects language development early on in life, and may continue to affect language development with increasing age.

In addition, 37% of parents/caregivers felt their child, adolescent or adult with Down syndrome had better communicative abilities compared to peers with Down syndrome of similar ages. This may reflect the good pragmatic skills and sociability of individuals with Down syndrome. The relatively stronger pragmatic and social skills of individuals with Down syndrome may be compensating for their weaker skills in other domains of speech and language, leaving to overestimation of overall communicative abilities by their parents/caregivers. It is consistent with findings regarding pragmatic ability in Roberts et al.'s (2007) and Abbeduto et al.'s (2007) reviews, where individuals with Down syndrome have good conversation skills and have strengths in gesture use. The percentage of individuals who were perceived to have better communicative abilities may be low because Down syndrome has a variable phenotype that ranges across a spectrum (Roper and Reeves 2006; Roberts et al. 2007).

In summary, *the survey data as well as the case studies show a high prevalence of sleep problems in individuals with Down syndrome regardless of the status of their language abilities, which may be a subjective view of parents/caregivers. The nature of the survey study does not allow for causality-related comments. To learn further about speech, language and behaviours of children with Down syndrome, I observed three therapy sessions and four weeks of summer school at the DSRF. My observations and summer study participation taught me that language and behaviour are important issues that are affected by many factors. The question is to what degree can the SLP help? As a first step, a prospective observational study investigating speech and language development as well as sleep problems using structured behavioural observations and video recordings from different perspectives may help find the answer. I present the protocol in the next chapter.*

2

PROTOCOL

2.1 Study Framework

The long-term goal is to identify how sleep quality within the first two years of life affects language development in children with Down syndrome. The proposed protocol supports this goal. The study is built on serial video recordings: sleep quality over night, alertness vs. tiredness at breakfast, receptive and expressive language as well as interactions at breakfast will be recorded and investigated.

2.1.1 Hypothesis

I hypothesize that the severity of speech and language difficulties in children with Down syndrome correlates with the severity of their sleep problems.

2.1.2 Research question

The overarching research question at the core of my interest is "*how do speech and language development interact with sleep problems?*" If an association can be demonstrated, we may be able to identify sleep as a reliable predictor of language, thus brain development. This would enable early identification and intervention to treat sleep problems starting at a young age to ultimately improve language abilities and development in children with Down syndrome.

2.2 Methodology

The study is based on close parent-professional collaboration and builds on video recordings obtained and provided by parents and analyzed by the professional: standardized serial sleep and wake behaviour observations will be recorded with a portable video system. As I plan on becoming an SLP in the future, I focus on what I can do myself as an SLP student. I am using the methodology I have learned over the last three summers at the Sleep/Wake-Behaviours Clinic and Research Lab. We must develop a shared language with parents to describe and discuss observations. By partnering with parents, we consider different perspectives and can make insights into what is underlying their child's behaviours.

At the Sleep/Wake-Behaviours Clinic and Research Lab, the disruptive and/or challenging behaviours of children with neurodevelopmental and mental health conditions are investigated with the Listen, Observe, Explore, Describe [LOED-] methodology (Ipsiroglu 2016). *Listening* (L) happens through the entire process of history taking and information gathering (Ipsiroglu et al. 2013). *Observing* (O) wake behaviours of the child and accompanying parents also happens through the entire process. Most importantly, the gathered data is *explored* (E) and symptoms are discussed and negotiated together (Ipsiroglu et al. 2016, 2017). *Describing* (D) the observation-based data and synthesizing it with parental descriptions and previous observations reinforces exploration. The development of an empathy-based approach, the LOED-methodology, supports and catalyzes "putting ourselves into the shoes of the children" (Frank 1985), and may equip us to better understand the dimensions of so-called disruptive and challenging behaviours (Ipsiroglu 2016). Video recordings, selected as the tool for applying the LOED-methodology, facilitates both the qualitative and quantitative analyses of a child's sleep/wake-behaviours as well as speech and language development. As video recordings provide a reproducible setting for various analysis

concepts, they also allow for equal consideration of the parent's/caregiver's and clinician's perspectives.

Methodological information: Patient population (eligibility). English-learning children with Down syndrome age 5 years or younger; there will be multiple entry points below the age of 5 years. The children's parents/caregivers will be English-speakers; children may be receiving speech therapy. **Length of study.** Study intake will occur when parents/caregivers agree with the consent form and volunteer to participate in the study. The study will last for one year in total. **Assessment concept.** Children are assessed at three time points to track individual development: initial assessment at study intake, follow-up assessment at six months, and follow-up assessment at twelve months. One week is monitored at each time point because of individual day-to-day and night-to-night variability. Video recordings provided by parents/caregivers are analyzed following adapted protocols. Parents/caregivers additionally identify periods of interest (POIs) warranting further investigation. This in-series and longitudinal data collection allows us to pool data blocks, which will most likely monitor interactions between sleep quality and the ability to learn in the subsequent morning to unveil individual speech and language development.

2.2.1 History taking: How to gain information?

Physicians gain main information from their patients by asking open-ended questions (Takemura et al. 2005). Surveys and questionnaires are frequently used as they provide a structured setting and format. In this subchapter, I describe, among various assessment approaches, three clinical practice-oriented concepts, which I reviewed given their use at the Sleep/Wake-Behaviours Clinic and Research Lab, and will justify my choice.

(1) **Children's Sleep Habits Questionnaire (CSHQ; Owens et al. 2000).** The CSHQ is a parent-report measure of sleep problems classified into eight domains covering problems in bedtime resistance, sleep disorders, and excessive daytime sleepiness. Parents are asked to recall the recent typical week of the child's life when answering questions. Example questions include: Child goes to bed at the same time at night. Child sleeps the right amount. Child snores loudly. Child awakes once during the night. Child has difficulty getting out of bed in the morning. Child seems tired. Responses are numerically summed to provide subscale and total scores. Responses are summed to create a total score. A score of over 41 indicates a possible sleep disorder.

(2) **The BEARS concept (Owens and Dalzell 2005).** "BEARS" is an acronym that stands for Bedtime, Excessive daytime sleepiness, Awakenings, Routines, Sleep-disordered breathing. It is designed to elicit comments from parents. The BEARS concept allows for open-ended, exploratory discussion and conversation with the parents and families and, as an explorative tool, has been implemented at the Sleep/Wake-Behaviours Clinic and Research Lab. Example questions include: What is your child's bedtime routine? Which strategies work and which do not? Does your child wake up during the night, and what do they do when they wake up? How does your child behave during the day? What kinds of daytime activities does your child participate in?

(3) **Sleep Disturbance Scale for Children (SDSC; Bruni et al. 1996).** The SDSC is a standardized and validated 27-item scale designed to categorize sleep disorders in children based on six subscales covering a spectrum of sleep disorders. The

SDSC includes all elements of the BEARS concept. Parents are asked to recall the past six months of their child's life when answering questions about their child's sleep/wake rhythm and the frequency of behaviours. Example statements include: The child goes to bed reluctantly. The child gasps for breath or is unable to breathe during sleep. You (the parent) have observed the child sleepwalking. The child startles or 'jerks' parts of the body while falling asleep. The child awakes in the morning feeling tired. The child sweats excessively during the night. Responses are numerically summed to provide six factor scores and a total score. A score of over 36 indicates a possible sleep disorder.

Discussion of methods: CHSQ. Advantage. The CSHQ has been used 147 times in published research studies; thus, we can assume that it is used more frequently clinically. Disadvantages. The CSHQ fulfills only five of the eleven fundamental operational principles of instrument development (Spruyt and Gozal 2011). When four subscales of the CSHQ (sleep onset delay, sleep duration, night wakings, and sleep disordered breathing) were validated against sleep studies (which are considered the gold-standard measure of sleep), the subscales did not correlate with the corresponding sleep study variables (Markovich et al. 2015). **The BEARS concept.** Advantage. The BEARS concept is open-ended and allows parents/caregivers to comment on a variety of factors related to their child's sleep and sleep quality. Disadvantages. The BEARS concept is not a validated questionnaire nor tested for reliability (Luginbuehl and Kohler 2009); thus, the concept is used in a discussion-based environment to identify specific sleep problems. **SDSC.** Advantages. The SDSC was developed following all eleven fundamental principles of instrument development (Spruyt and Gozal 2011). It was normed on over 1000 children including 147 with sleep disorders (Bruni et al. 1996). The subscales fit the diagnostic classification of sleep

and arousal disorders (Spruyt and Gozal 2011). The SDSC has good diagnostic accuracy; the sleep breathing disorder subscale has been validated with sleep study-confirmed sleep-disordered breathing (Ferreira et al. 2009). Time to complete the questionnaire is approximately ten minutes (Bruni et al. 1996). The SDSC is also a multidimensional measure (Lewandowski et al. 2011) that provides a standardized scale for the assessment of sleep problems. Disadvantages. The SDSC is not open-ended and does not account for individuality (de Souza Vilela et al. 2016).

In summary, *the BEARS concept explores parental view and allows for qualitative answers while the SDSC, as a validated questionnaire, will capture 'medically' sleep problems. The BEARS concept and the SDSC complement each other, taking two approaches (open-ended vs. standardized) to understanding the child's behaviours. Both together will help to develop a shared language for focusing on sleep disorders and their interactions with disruptive daytime behaviours (including language and speech) caused by sleep problems after the initial assessment and follow-ups. The BEARS concept and SDSC will be completed once at each time point.*

2.2.2 The role of the family in gathering information

History taking should be complemented with a tool aimed at collecting direct observations of the child's sleep- and wake-behaviours over time. In this subchapter, I describe the sleep log/diary.

The sleep log/diary measures the sleep/wake patterns of the child. On the log, parents/caregivers are asked to record the times when their child goes to bed, when their child is actually asleep, and when their child wakes up in the morning. The sleep logs/diaries used at the Sleep/Wake-Behaviours Clinic and Research Lab have additional questions about the child's exercise, disruptive behaviours (e.g. a temper tantrum), medications, and snacking behaviour. There is space to encourage parents to mention or report the major daytime and nighttime

challenges that are perceived by parents. Parents/caregivers are asked to comment on any of the above and are also asked to give a rating of their child's sleep quality for the night. In studies with populations exceeding current expertise, e.g. Down syndrome, other specific questions can also be added to the sleep log/diary to generate additional open-ended, exploratory questions.

Discussion of method: Advantages. The sleep log/diary collects specific information on the sleep/wake pattern and additional questions can be added to elicit further descriptions. It also enables parents/caregivers to record their own observations during the day and night. Disadvantage. The data collected from the sleep log/diary is limited to parental/caregiver awareness of their child's behaviours (Sadeh 2015).

In summary, *the sleep log/diary supports parents/caregivers to record their observations and is complementary with the previously mentioned assessment tools for use at home. This provides further insight into the happenings at home. The sleep log/diary will be completed for one week of nighttime behaviour monitoring at each assessment time point.*

2.2.3 How to objectively monitor nighttime behaviours?

Sleep is a complex behaviour that can be assessed at different levels and depths (Sadeh 2015). Different methods are available depending on the points of interest and depth of assessment (Sadeh 2015). In this subchapter, I describe two routine methods for assessing sleep quality and sleep/wake-behaviours in children with Down syndrome.

(1) **Actigraphy.** Actigraphy is a watch-like device used for measuring sleep/wake periods to assess sleep fragmentation and quality. It continuously monitors body movements (Sadeh 2015). The watch-like device can be fitted to the ankle for infants and younger children or to the wrist for older children and adults (Sadeh

2015). The raw activity data collected is scored using a computer algorithm and then transformed to sleep/wake scores for further analysis of sleep/wake patterns (Sadeh 2011).

(2) **Home video recordings (Ipsiroglu et al. 2015).** Home video recordings are performed in the natural and familiar setting of the child's home. Parents/caregivers are provided with an infrared camera and netbook, and set up the system facing the child's bed to record the child's behaviours from bedtime the night before to wake-up the morning after. The video data is analyzed according to a standard analysis pipeline and protocol (Ipsiroglu et al. 2015): (1) Key features are first identified in qualitative analysis including patient behaviours and bedtime setting. (2) The video is annotated in an overview to explore and describe the content where key features that may affect sleep are identified, and notes are made about the various periods of sleep. (3) A second viewing allows for more detailed notes about movement patterns and behaviours. (4) The final viewing is for listening and in-depth descriptions of patient-generated sounds while viewing the video in real-time. (5) Further quantitative analysis is performed to analyze movement in the video.

Discussion of methods: Actigraphy. Advantages. No installation of equipment is required and actigraph-measured sleep can be analyzed using a computer algorithm (Sadeh 2015). Disadvantages. Actigraphy may not have a high specificity in children with complex developmental disabilities including children with Down syndrome (Sadeh 2011; Meltzer et al. 2012). Technical parameters, such as device and algorithm selection, also pose challenges when analyzing actigraphy data (Sadeh 2015). As the actigraph is placed on the participant's wrist, behaviours that help characterize sleep, such as facial expressions, head/neck position,

parasomnias, and restlessness, e.g. due to periodic limb movements and twitching, are not recorded. In addition, other behaviours, which are important and affect sleep, such as emotions, anxiety and level of distress including interactions with parents/caregivers, are missed (Ipsiroglu et al. 2015). **Home video recording.** Advantages. Home video recordings captures the entire bedtime and sleep situation overnight in a natural setting at the child's home as well as all observable behaviours, including interactions with caregivers, child movements and awakenings. A standard protocol exists for analysis of video data (Ipsiroglu et al. 2015). Disadvantages. It may be perceived as time consuming for parents/caregivers to install the equipment, and some parents may have privacy concerns (Sadeh 2015).

In summary, *home video recordings facilitate the observation and description of sleep/wake-behaviours. Home video recordings are used to monitor sleep patterns during the night, from bedtime to final wake-up. One week is monitored at each assessment time point due to individual night-to-night variability. Behaviours will be described and analyzed according to a standard pipeline and protocol. All descriptions are discussed with parents/caregivers to develop a shared language focusing on sleep problems.*

2.2.4 How to monitor morning behaviours?

Behaviours like EDS or simply being tired in the morning reflect the quality of sleep received the night before, and are thus important to describe. Based on this understanding, I am suggesting monitoring behaviours during breakfast. Of special interest are: vigilance, interactions with parents/caregivers, and receptive and expressive language abilities (oromotor skills). Behaviours are recorded and then the recordings are analyzed for various themes among behaviours.

The current strategy for recording language in real-time is **Language ENvironment**

Analysis (LENA; LENA Research Foundation), developed by the LENA Research Foundation in Boulder, Colorado, USA. LENA uses a recorder worn by the child in a vest to record up to 16 hours of speech and speech-like sounds. A computer algorithm for automated language analysis separates recorded speech and speech-like sounds from environmental noise, and then identifies the speakers in the sample as adult male, adult female or child wearing the recorder. The algorithm analyzes the sample for each speaker for different points of interest, such as number of child vocalizations, number of parent/caregiver utterances, and number of conversational turns. The advantage of using LENA is that it does not require extensive setup or installation of equipment. Disadvantages are that the recorder setup may provoke noncompliance in participating children. The advantage for using LENA in children with Down syndrome is that the children are assessed in the context of their natural environment, where their speech is recorded in real-time as they engage throughout the day. Disadvantages for using the system in children with Down syndrome are: (1) LENA has not been validated for use in children with Down syndrome, although it was used in a study by Edgin et al. (2015). (2) LENA does not record nonverbal communication. The LENA system does not account for gesture production, which is a strength in children with Down syndrome (Abbeduto et al. 2007), nor signs that are produced. (3) Parent/caregiver interactions are limited to conversations. Thus, a different system must be used to characterize morning behaviours.

Video recordings, as suggested for this study, may enable us to capture the child's behaviours during the breakfast environment. Advantages of using a video system are that tiredness, EDS behaviours (e.g. inattention to others, vigilance), as well as interactions are directly observed and can be described. The video recording also facilitates recording the child's speech, gestures and signs, which can be transcribed for further analysis. The disadvantages are that

parents/caregivers and the participating children have privacy concerns or feel uncomfortable being recorded during breakfast as their face, body movements, and actions are recorded. Additional issues are that the quality of the audio and video must be high definition to allow for clear observation of behaviours and accurate transcription of speech. Data storage and transfer are also more complex than with the LENA system, and analyses may be time consuming. Though there are possible drawbacks, which need to be solved, video recordings may still be an optimal method for generating reproducible observations. I describe the procedure below.

Setup and concept of Social Stories™. To increase the child's comfortableness around the camera, the camera is introduced using a Social Story™. Social Stories™ were originally developed by Carol Gray, an American teacher who worked with children with autism spectrum disorder. Social Stories™ are used for a variety of purposes, including to introduce changes and new routines, to teach social skills and to explain the behaviours of others (Gray and Garand 1993). They are written from the child's perspective (Gray and Garand 1993) and often are accompanied by pictures depicting each sentence. Three main sentence types are used in Social Stories™ (Gray and Garand 1993): (1) *Descriptive sentences.* Descriptive sentences accurately describe the social situation. These sentences state what others do in a neutral tone. (2) *Perspective sentences.* Perspective sentences describe the responses of others to the same social situation. These sentences help the child understand different reactions to the social situation. (3) *Directive sentences.* Directive sentences state desired behavioural responses. These sentences describe what is appropriate for the child to do to succeed in the situation. There should be fewer directive sentences to allow the child to determine their own response to the situation. The Social Story™ is created and developed with parents/caregivers, and is unique to each child (Gray and Garand 1993). An example story (**Table 4**) is provided below for "Taylor", a child with Down syndrome whom I

observed at the DSRF. The story introduces the camera used to make video recordings during his morning breakfasts. In this example, the story is read to "Taylor" while he follows the pictures and because he cannot yet read. Note that "Rachel" is the pseudonym for "Taylor's" SLP.

Table 4: Example Social Story™ for "Taylor". The title is: "Helping Rachel learn about how I talk at home."

Picture	Sentence	Sentence Type
The front door to the DSRF.	We visit the DSRF each week to see my speech therapist, Rachel.	Descriptive
Rachel waving 'hello'.	I practice my speech and language skills with Rachel every week.	Descriptive
Rachel teaching Taylor to use his *I want…* sentence strip.	Rachel teaches me to use my words and my *I want…* sentence strip to talk to others.	Descriptive
Rachel working with Taylor.	Rachel sees me for 45 minutes each week because other kids also need help.	Descriptive
Rachel with her "thinking" face.	Rachel wants to learn more about how I talk so she can help me more.	Descriptive
Rachel with the camera set up.	Rachel has an idea. Rachel gave us a camera to record me during breakfast at home so she can see how I talk at home.	Descriptive
Taylor and his teddy bear.	The camera on one day records me talking to my teddy bear.	Descriptive
Family eating breakfast in the morning.	The camera on another day records me talking at breakfast.	Descriptive
Rachel and Taylor posed together for a photo.	This helps Rachel learn more about how I talk at home.	Descriptive

Table 4 presents an example Social Story™ for "Taylor".

This story is initially read with the parent/caregiver and afterwards, the child reads the story on their own (Gray and Garand 1993). The child then reads the story once per day to solidify learning of the desired behaviour. Comprehension is checked using role playing (Gray and Garand 1993). Using a story-based introduction of the camera may facilitate a smooth transition into the recording procedure. During the recording period, as the parents/caregivers are recording by themselves at home, the camera is set up in the same (convenient) location in the family's dining area for each morning's recording.

Data. Breakfast time varies in length from family to family and child to child, so it may be difficult to analyze the recordings in full, particularly if the recordings are lengthy. The analysis procedure is adapted from Ipsiroglu et al. (2015): The video recording is first viewed at 8 times normal speed to note key features, obtain a general 'feel' for behaviours and to identify POIs. POIs are additionally identified by parents/caregivers (e.g. for periods that warrant further attention from the parent/caregiver perspective). Next, the video is reviewed at 4 times normal speed to make more detailed notes and to verify previously identified POIs. Finally, the video is reviewed in real time to take in-depth notes. The video recordings in this study protocol are annotated and analyzed for vigilance, interactions and oromotor skills (**Tables 5-7**).

Analysis of vigilance. Vigilance is the ability to stay concentrated for a period of time (Ipsiroglu et al. 2017). The analysis procedure from Ipsiroglu et al. (2017) is adapted for this study protocol. Several factors are described and ranked to assess vigilance, presented in **Table 5** (Ipsiroglu et al. 2017): body posture, sleepiness, attention, facial expressions. In addition to ranking behaviours, videos are analyzed quantitatively with optical flow analysis (Barbosa et al. 2008), a software that extracts and analyzes 2D motion from consecutive frames in a video recording. The optical flow analysis allows us to understand how motion in a visual scene is perceived (Latif et al. 2014). Pixel brightness is used to measure motion amplitude of moving structures (e.g. head, face, torso, arms, legs) over time (Barbosa et al. 2008; Ipsiroglu et al. 2015), which helps characterize movement patterns.

Table 5: Analysis of vigilance (Ipsiroglu et al. 2017).

Factors	Descriptions

Body posture	• Attributes or characteristics
	  o Are they relaxed or do they have increased tension?
	• Child's position in space:
	  o How is the child sitting at the table?
	  o Do they have their head down, are they slouching or are they sitting with their back straight?

Sleepiness	• Distinguish between hypo-vigilant, hyper-vigilant and vigilant
	  o Hypo-vigilant: drowsy, tired, yawning, rubbing eyes/face
	  o Hyper-vigilant: hyperactive, nervous, anxious, increased tension
	  o Vigilant: awake, does not display any of the above for hypo- or hyper-vigilant

Attention	• Distinguish between hyper-focused, hypo-focused, and receptiveness to others
	  o Is attention selective or is it divided/alternating?
	  o Is attention sustained?
	  o Where is attention directed: towards self or others?
	  o Can they respond to others? How distracted are they?

Facial expressions	• Facial features
	  o What emotions are they expressing?
	  o Are they frowning, scowling, have their eyebrows raised?
	• Rate the variability of smiles on a 5-point scale: 1 = oversized/tense smile with abrupt onset and offset, 5 = fine/variable smile with smooth onset and offset

Table 5 presents the analysis procedure for assessing vigilance from the video recording.

Analysis of interactions. There are three types of interactions (Ipsiroglu et al. 2017): social interaction, with others including parents/caregivers and other family; self-interaction; and object interaction. The analysis procedure from Ipsiroglu et al. (2017) is adapted for this study protocol, presented in **Table 6**: interactions are described for tone of voice, quality, as well as modality. An optical flow analysis is performed to analyze movement coordination (e.g. facial expressions, postures, speech rates) between the child and their parent/caregiver during a social interaction (Latif et al. 2014). Correlated movements indicate coordination and rapport between speakers.

Table 6: Analysis of interactions (Ipsiroglu et al. 2017).

Factors	Descriptions

Tone of voice	• Speech etiquette
	  ○ In what manner does the child communicate with others (e.g. happy, relaxed voice; yelling and screaming; angry voice)?
	  ○ Is the child speaking with normal volume, or are they quiet or loud?
	  ○ If signing, at what intensity are the signs made?

Quality	• Kind of interactions, e.g. cooperative, competitive, commenting, playing • Directionality: one-way or reciprocal • Analyze for frequency and duration
Modality	• Distinguish between verbal and nonverbal (e.g. signs, gestures, eye gaze) • Analyze for frequency and duration

Table 6 presents the analysis procedure for assessing interactions from the video recording.

Analysis of oromotor skills. A special note is that speech abilities in typically developing children are interconnected with oromotor skills (Tittnich et al. 1990); this is also true for children with Down syndrome (Cleland et al. 2010). But to what degree oromotor skills are affected by sleep has not been investigated. Video recordings may provide insight. The analysis procedure from Ipsiroglu et al. (2017) is adapted for this study protocol, presented in **Table 7**. There is currently no standard for assessing oromotor (speech and non-speech) skills in children with Down syndrome, but an assessment is being developed in the research setting (Rupela et al. 2016). Thus, for this study, I suggest that oromotor skills are assessed for muscle tension and articulation of sounds using the video recordings. An optical flow analysis is then performed to quantify head and facial movements that occur in coordination with the child's speech (Barbosa et al. 2008).

Table 7: Analysis of oromotor skills (Ipsiroglu et al. 2017).	
Factors	**Descriptions**
Muscle tension	• Attributes or characteristics o Are the orofacial muscles tensed, relaxed, or is the mouth wide open? o How does the child breathe - through their nose or their mouth?
Articulation	• Clarity of speech o Are the facial movements coordinated during speech? o Is the child's articulation slow and do speech sounds blend together?

Table 7 presents the analysis procedure for assessing oromotor skills from the video recording.

In summary, *morning behaviours reflect the quality of sleep from the night before. It is important to describe vigilance, interactions with parents/caregivers and oromotor skills as each may be affected by sleep. These three behaviours of special interest are not available from the LENA system. Based on the above discussion, video recordings may be optimal for facilitating observations at breakfast. Video recordings and behaviours are analyzed using adapted protocols from Ipsiroglu et al. (2015) and Ipsiroglu et al. (2017), respectively. Optical flow analyses are performed for quantitative analysis to complement qualitative descriptions of behaviours. One week is monitored at each assessment time point due to individual morning-to-morning variability. All descriptions and analyses from the video recordings are discussed with parents/caregivers to validate the observations, to solicit further observations from the parents/caregivers and to develop a shared language for discussion of morning behaviours.*

2.2.5 How to investigate speech and language abilities?

Language abilities must be monitored to track individual progress and development in this domain. Speech-language pathologists use a variety of tools (standardized and non-standardized) to assess receptive and expressive language abilities (Crais 2011). In this subchapter, I describe four

methods to assess language and speech that could be used in this study protocol.

(1) **The MacArthur-Bates Communicative Development Inventory (CDI; Fenson et al. 2007).** The CDI comes in two forms for two different age groups and is a measure of the child's vocabulary. Parents/caregivers are asked to indicate which words their child uses/produces and understands. Questions on the CDI also ask about word endings (grammatical properties), how the child uses words and the complexity of the child's sentences.

(2) **The Clinical Evaluation of Language Fundamentals Preschool-2 (CELF-P2; Wiig et al. 2004).** The CELF-P2 is a standardized assessment designed to detect the presence and severity of a language disorder in three- to seven-year-old children and to identify which domain of language it affects (Wiig et al. 2004). The test comprises several levels. For the most basic level (called the Core Language Score), children are asked to point to pictured objects, complete sentences and label pictures. The Core Language Score represents the child's overall language abilities.

(3) **The Structured Photographic Articulation Test II (SPAT-II; Dawson and Tattersall 2001).** The SPAT-II is an articulation assessment designed to assess consonants in the English language in all positions of the word, including consonant blends and common phonological processes (Dawson and Tattersall 2001). It is a story-based assessment that features a puppy in silly situations, and children are asked to name pictured objects or actions.

(4) **The Language Use Inventory (LUI; O'Neill 2007).** The LUI is a standardized parent report measure designed to assess the pragmatic language abilities of young children (O'Neill 2007). Parents/caregivers answer questions on 14 subscales

covering different aspects of pragmatic language. Questions ask about the child's use of gestures, words and longer sentences (O'Neill 2007) in daily life with various communication partners for a variety of purposes (Pesco and O'Neill 2012). Example questions are: Does your child point to something wanted? Does your child request for help with difficult tasks? Does your child ask for a turn? Does your child tell jokes?

Discussion of methods: CDI. Advantages. Validity of the CDI for parents of children with Down syndrome was established by Miller et al. (1995). Since then, the CDI has been used in several research studies focusing on children with Down syndrome (Singer Harris et al. 1997; Mervis and Robinson 2000; Vicari et al. 2000; Bird et al. 2005; Galeote et al. 2008, 2011; Zampini and D'Odorico 2009, 2013; Vanvuchelen et al. 2011; Foster-Cohen and van Bysterveldt 2016). The CDI has been adapted for 69 languages (CDI Advisory Board 2015); a Spanish adaptation specific to children with Down syndrome was also developed (Galeote et al. 2016). Thus, we assume that the CDI is frequently used by clinicians. Disadvantages. The CDI focuses on vocabulary and grammar, with few questions on other domains of language (e.g. phonology, pragmatics). The CDI may not reflect the complete language skills of children with Down syndrome given their weaknesses in vocabulary and expressive language (Abbeduto et al. 2007; Roberts et al. 2007). **CELF-P2**. Advantages. The CELF-P2 assessment uses a picture book to engage the child and does not require many props. The stimuli and tasks are school-focused. Responses to test stimuli (e.g. enthusiasm, attention, boredom, frustration) can be recorded using video for qualitative analysis (Driscoll-Davies 2010). The CELF-P2 takes 15-20 minutes to administer for the Core Language Score. Disadvantage. The CELF-P2 (UK version) has been used in only one published research study focusing on children with Down syndrome (Cleland et al.

2010), but in clinical practice, the CELF-P2 is used by speech-language pathologists at the Down Syndrome Research Foundation (Burnaby, Canada) for clients as required (Susan Fawcett, 2016, personal communication). **SPAT-II.** Advantages. The SPAT-II uses an animal in different situations as the stimuli for the assessment; this may hold the child's attention during the 30-minute test administration. The test words are modern words that children learn through daily experiences. Disadvantages. The SPAT-II has not been used in published research studies focusing on children with Down syndrome or with global developmental delay, but in clinical practice, it is used by speech-language pathologists at the Down Syndrome Research Foundation (Burnaby, Canada) for clients as required (Susan Fawcett, 2016, personal communication). Children may become distracted by the pictures and not participate in the assessment. **LUI.** Advantages. The LUI has been used in research studies focusing on children with Down syndrome (Schutz 2014; Foster-Cohen and van Bysterveldt 2016). The LUI is not limited to vocabulary or grammatical skills; it also accounts for nonverbal and gestural aspects of communication (O'Neill 2007). The LUI has a high sensitivity (true positive rate) of 81% and a high specificity (true negative rate) of 93% (Pesco and O'Neill 2012) for differentiating those with deficits compared to those without. The inventory was normed on a large community sample of Canadian children (O'Neill 2009) and can be completed in 20-30 minutes. Disadvantage. The LUI places an emphasis on language use (i.e. pragmatics) and may not fully capture grammatical skills and knowledge of words or semantics.

In summary, *none of the four tools reviewed in this subchapter are sufficient on their own to investigate individual speech and language abilities of children with Down syndrome because none captures speech and all domains of language in the assessment. Using the tools in combination may allow us to assess each domain and the data can be pooled to generate a profile. While it is useful to have a vocabulary measure, the CDI focuses on expressive language, which*

is a weakness in children with Down syndrome. Thus, the CELF-P2 is used to assess vocabulary in combination with syntactic abilities. The SPAT-II is used to assess speech abilities and phonology. The LUI, as a parent/caregiver tool, is used to assess pragmatic language. The three tools complement each other and the data gathered from the video recordings at breakfast. The CELF-P2, SPAT-II and LUI are each completed once at each assessment time point. All three together will help develop a shared language with parents/caregivers, and provide equal consideration of the parent's/caregiver's and clinician's perspectives.

3

GENERAL DISCUSSION

My literature review taught me that research on speech, language development and sleep problems in children with Down syndrome has focused on characterizing each area separately. To what degree sleep problems affect speech and language development has not yet been fully investigated. Edgin et al. (2015) started the research. Expanding their concept, I am suggesting the use of structured behavioural observations and video recordings to monitor speech, language development and sleep problems. The justification is based on observations in typically developing children (Rosen et al. 2002, 2004; Sadeh et al. 2002; Steenari et al. 2003; Gottlieb et al. 2004; Buckhalt et al. 2007; Seicean et al. 2007; Ievers-Landis et al. 2008; Anderson et al. 2009; Goodlin-Jones et al. 2009; Hall et al. 2012; Ross et al. 2012; Vasak et al. 2015; Cespedes et al. 2016) and in children with Down syndrome (Chen et al. 2013; Esbensen 2016; Lukowski and Milojevich 2017). The high co-occurrence of language difficulties and sleep problems in children with Down syndrome, as inferred from the literature (Chapman et al. 1991, 1998; Abbeduto et al. 2007; Roberts et al. 2007; Cleland et al. 2010; Stores and Stores 2013; Esbensen 2016) and as revealed by the Down Syndrome Needs Survey, call to question to what degree they are correlated. In this chapter, I discuss how the proposed study protocol will answer my central research question and detail some limitations of the protocol.

3.1 Discussion of research question

To answer the research question "*how do speech and language development interact with sleep*

problems?", I must first identify the relevant points of interest:

(1) **History taking.** Qualitative descriptions from the BEARS concept and parent/caregiver observations from the sleep log/diary are differentiated to highlight the main themes in behavioural patterns in the daytime, nighttime, as well as in transitions. The sleep log/diary captures asleep, awake and vigilance and mood-related information, such as time spent asleep as observed by parent, time spent awake, number of disruptive episodes, and time spent exercising.

The qualitative descriptions from the BEARS concept will be compared with the quantitative SDSC scoring (Bruni et al. 1996), and allow for understanding the range/dimension of descriptions with a validated standardized tool. The five BEARS domains can be compared directly with the six SDSC subscales. Descriptive statistics (frequencies, percentages and averages) are calculated for SDSC subscales and total scoring.

(2) **Home video recordings.** The home video recordings are quantified for the following points of interest (Ipsiroglu et al. 2015): *went to bed*, *lights out*, *sleep onset* (transition from wake to sleep), *sleep latency* (total time between going to bed and falling asleep), *wake time* (total time in minutes that is scored awake occurring between sleep onset and final wake-up), *total sleep period* (period of time measured from sleep onset to final awakening; equal to total sleep period less movement and awake time), *total sleep time* (amount of actual sleep time in a sleep period), *sleep efficiency* (proportion of sleep in the period potentially filled by sleep; ratio of total sleep time to time in bed), *restful sleep*, and *restless sleep* (persistent or recurring body movements, arousals and/or brief awakenings in the course of

sleep). This quantitative analysis of nighttime and sleep behaviours complements the qualitative measures of history taking and sleep log/diary. Night-to-night variability in the children's sleep patterns is addressed by monitoring one week of sleep and pooling data across several nights.

(3) **Video recordings of morning behaviours.** Video recordings are quantified for three factors following an adapted protocol (Ipsiroglu et al. 2017): vigilance, interactions and oromotor skills (described above). Descriptions are then differentiated to highlight recurring themes. Transcribed speech, gestures and signs are analyzed for average sentence length and vocabulary. Morning-to-morning variability is addressed by monitoring one week of breakfasts and pooling data across several mornings.

(4) **Speech and language investigation.** The CELF-P2 and SPAT-II provide raw scores and total scores. For the SPAT-II, other variables, such as the percentage of consonants correct, consonant and vowel inventory sizes, and total sound repertoire size, are additionally analyzed. The LUI provides subscale and total scores.

Then, as a first step, descriptive analyses are used to describe behaviours. The mean and median for each point of interest is calculated. Results are examined for trends (e.g. if higher means on measures of speech and language development are associated with higher means on measures of sleep quality). This will most likely provide insight into possible interactions.

3.2 Discussion of methodology

The methodology suggested in this thesis provides a way of *listening*, *observing*, *exploring* and *describing* the nighttime and daytime behaviours of children with Down syndrome. To facilitate

individual tracking of how speech and language development interact with sleep problems, the proposed study protocol contains a longitudinal element: the same individuals are measured at three time points to describe individual development. An alternative to this is a series of cross-sectional assessments, but this would not allow for individualized monitoring. Cross-sectional assessments measure different individuals at each time point. Thus, the longitudinal element is selected.

My role as an SLP student. The proposed study protocol focuses on what I can do myself, as a future SLP student. This protocol will form the basis of my Master's studies and thesis to build on the concept of possible interactions between speech, language and sleep problems. This proposed protocol also fits with my future role in the clinic as a facilitator of a therapeutic setting to understand clients, their families and their communication needs. The real life setting we have chosen is the breakfast situation and allows assessment of vigilance as a standard for sleep quality, interactions of parent/child, as well as to what degree oromotor skills might be influenced by sleep, or present the underlying severity of the Down syndrome phenotype. The suggested tools support therapeutic conversations by allowing the discussion of structured observations. This will optimize my understanding as a facilitator of a therapeutic setting and allow me to gain a deeper insight in the main points of interest. There is reciprocity in added value. Parents/caregivers are also getting a better understanding of their communication needs and are enabled to optimize not only their routines, but, by reviewing the video sequences, reflect and possibly optimize their interactions. Video recordings allow for the consideration of multiple perspectives on the child's behaviours through viewings by myself, parents/caregivers and (if necessary) by other clinicians. This enables not only the development of a shared language between caregivers and myself for discussing sleep/wake-behaviours, but also the deeper investigation of unexplored inter-connections as

described in my hypothesis.

Limitations. There are two main limitations to the proposed study protocol. *First,* demographic factors may influence the results. For example, children from families with lower socioeconomic status develop language more slowly compared to children from families with higher status, possibly because they receive lower quality language input from their parents/caregivers (Hoff and Tian 2005). In addition, child birth order is also positively correlated with language development: firstborns tend to receive more speech from parents than later-borns, and thus develop language with fewer delays (Huttenlocher et al. 2010). Though this is an important limitation, I believe that the limited number of participants and longitudinal character of the proposed study is giving us different data. Socioeconomic status may play a role and it may be reproducible in our study, but my assumption is that it would be represented mainly through interactions and possibly tiredness/exhaustion of the parents. *Second,* children with Down syndrome have characteristic but uneven phenotypes, resulting in differences in great variability in development. This may be perceived as a limitation, but it is an advantage of the study. To what degree (uneven) developmental progress is affected by sleep problems may be solved because analyses of sleep/wake-behaviours are individualized for each participant to account for each individual phenotype.

In summary, *the LOED-methodology suggested in this study provides qualitative and quantitative data on children's sleep/wake-behaviours. The selected methods complement each other to assess behaviours using a rounded, comprehensive methodology. This proposed study protocol, as a first step, builds on a previous concept by Edgin et al. (2015). It exceeds previous work through its comprehensive, clinical and longitudinal elements, and may provide insight into how speech and language development interact with sleep problems as well as new information*

and ideas for further discussions.

4

CONCLUSION

By understanding how speech and language development interact with sleep problems, we may be able to gain deeper insights into the effects of sleep on the developing brain. This may be best studied in various clinical models. I am suggesting monitoring *speech and language development* over three time periods with serial measures and combining it with sleep, vigilance and the interactions the subsequent morning in children with Down syndrome. As I plan on becoming an SLP in the future, I focus on the role of the SLP and to what degree SLPs can facilitate this therapeutic conversation with clients and their families. If an association between speech and language development and sleep problems can be demonstrated, this enables early intervention to improve the speech and language abilities of children with Down syndrome. This would improve their wellbeing and quality of life.

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