The Relation between Gestures and Stuttering in Individuals with Down syndrome

Abstract

Background: Evidence shows that neurotypical individuals who stutter use fewer gestures than those who do not stutter. Presently, no research exists about the interaction of stuttering and gestures in individuals with Down syndrome.

Method: Twenty-nine individuals with Down syndrome (7-19 years) of whom 16 stuttered and 13 spoke fluently and 20 neurotypical children (3-10 years) of whom 8 stuttered and 12 spoke fluently participated in this study. In spontaneous speech transcriptions, stuttering events and gestures were coded.

Results: Comparisons of gesture frequency during stuttered and fluent speech inside the Down syndrome and neurotypical group show that the Down syndrome group uses significantly more gestures during stuttered than during fluent speech while no significant difference is seen in the neurotypical group.

Conclusions: There is some preliminary evidence that individuals with Down syndrome try to compensate for their stuttering events, however, analyses on word level are necessary to confirm a successful compensation.

Keywords:

Down syndrome, Gestures, Stuttering, Gesture-Speech relationship

1. Introduction

1.1 Down syndrome

Down syndrome is the most occurring chromosomal disorder with a prevalence between 1 in 400 to 1 in 1500 births across different countries (Chisholm, 2018; Kazemi, Salehi, & Kheirollahi, 2016). Individuals with Down syndrome show greater impairments in their language development than would be expected based on their intellectual disability (Chapman, 1997; Næss, Lyster, Hulme, & Melby-Lervåg, 2011). Despite their problems with verbal communication, they show some strengths in their non-verbal communication, such as the use of gestures (Smith et al., 2017). There is evidence that individuals with Down syndrome are able to use gestures to express a meaning they cannot convey in spoken words (Lorang, Sterling, & Schroeder, 2018; Stefanini, Caselli, & Volterra, 2007; Stefanini, Recchia, & Caselli, 2008). Besides possible problems in language, which involve receptive and expressive vocabulary, syntax, and morphology, individuals with Down syndrome can also have difficulties in different areas of speech production (Bray, 2008; Kent & Vorperian, 2013; O' Leary, Lee, O'Toole, & Gibbon, 2020). Speech production involves the coordination of voice, articulation, prosody and fluency to make the correct production of consonants, vowels, phonemes and words (Cleland, Wood, Hardcastle, Wishart, & Timmins, 2010). For example, somebody who says 'I would like that a-a-a-apple' shows a speech (stuttering) problem but not a language problem. Conversely, somebody who says 'I yesterday an airplane gone on holiday' shows a language (syntax) problem but not a speech problem.

Currently, there is no evidence that individuals with Down syndrome use gestures to support their speech during stuttering problems as they do to support their language during language production problems. The present study takes a first look at how stuttering interacts with the gesture use of individuals with Down syndrome.

1.2 Stuttering

A specific speech problem in individuals with Down syndrome is stuttering (Devenny & Silverman, 1990; Eggers & Van Eerdenbrugh, 2018; Kent & Vorperian, 2013; Otto & Yairi, 1974; Preus, 1972; Van Borsel & Tetnowski, 2007). Stuttering is a disorder in the fluency of speech that is characterized by involuntary disfluencies such as sound, syllable and/or monosyllabic word repetitions (e.g. b-b-b-backpack; pe-pe-pe-pensil; we-we-we-we), sound prolongations (e.g. mmmmoney; niiiiice) and blocks (e.g. a…..apple), where the airflow is temporarily obstructed and the speaker cannot move forward (Ambrose & Yairi, 1999; Scott, 2017). These stuttering-like-disfluencies are separate from typical disfluencies such as polysyllabic whole-word repetitions (e.g. yesterday – yesterday), phrase repetitions (e.g. I will I will go away), phrase revisions (Dad said/Dad asked what we did) and interjections (e.g. uhm) that occur in the speech of both individuals who do and who do not stutter (Ambrose & Yairi, 1999).

Stuttering appears to have a high occurrence in individuals with Down syndrome with a mean prevalence around 30% (Devenny & Silverman, 1990; Eggers & Van Eerdenbrugh, 2018; Kent & Vorperian, 2013; Otto & Yairi, 1974; Preus, 1972; Van Borsel & Tetnowski, 2007), while in individuals with intellectual disability and in individuals with typical development it is respectively 5% and 1% (Van Borsel & Tetnowski, 2007; Yairi & Ambrose, 2013). Individuals often try to cope with these disruptions, resulting in secondary behaviours (Scott, 2017). Secondary behaviours are coping strategies that lose their fluency enhancing effects and become classically conditioned parts of the stuttering events that add to the stuttering severity. For example, someone who nods with every stuttering event. Information about stuttering therapies for individuals with Down syndrome is scarce. Bray (2015) mentioned that traditional therapies require certain abilities that are often too difficult for individuals with Down syndrome such as self-monitoring and active learning. However, although debated (Van Borsel & Tetnowski, 2007), stuttering can have a negative impact on the confidence and self-esteem of individuals with Down syndrome (Jackson, Cavenagh, & Clibbens, 2014), and should therefore be treated. Bray (2015) reported that parents often observe spontaneous coping strategies in their children with Down syndrome, such as slowing down their speech or substituting difficult words. She argued that these behaviours, which are seen as secondary behaviours, should be redefined in individuals with Down syndrome. Working with behaviours already present is less cognitive demanding and will aid with retention (Bray, 2015). Because gesture use is a strength in individuals with Down syndrome (Smith et al., 2017), this might be a behaviour that they use as a coping strategy for stuttering.

1.3 Gesture use

Gestures can be defined as spontaneously moving body parts during speech (Abner, Cooperrider, & Goldin-Meadow, 2015; Kendon, 2004). Gestures can be produced by the head, the hands, the shoulders, … (Vanderdonckt et al., 2019; Wagner, Malisz, & Kopp, 2014) but the present study focusses on the gestures produced by the hands. Spontaneous hand gestures have two main functions: a substantial (or referential) function or/and a pragmatic function (Kendon, 2004). Substantial gestures are related to the semantic content of an utterance, and they encompass *iconic gestures*, *metaphoric gestures*, and *deictic gestures* (McNeill, 2015). Iconic gestures represent concrete objects or actions, metaphoric gestures represent abstract meanings, and deictic gestures are pointing gestures. Gestures with a pragmatic function have a less direct relation to content. They are divided in pragmatic discourse gestures, also called beats gestures, and other pragmatic gestures. Beat gestures have no strict hand shapes and help the structure of the speech by highlighting words, controlling the parsing and following the rhythm of the utterance (Biau & Soto-Faraco, 2013; McClave, 1994; Prieto, Cravotta, Kushch, Rohrer, & Vilà-Giménez, 2018). Other pragmatic gestures involve pragmatic gestures with a modal, performative, or interpersonal function. A modal gesture shows the speaker's stance, a performative gesture indicates an interactional move and an interpersonal gesture is used for organizing the interaction. It is also possible that a gesture has both a substantial and pragmatic function, for example counting fingers while listing people is both a metaphoric representation of these people and an aid in structuring the speech (Cienki & Müller, 2008; Kendon, 2004; Tuite, 1993; Wagner et al., 2014). Spontaneous hand gestures are separate from adaptors, such as twirling one's hair, that fulfil no function in the communicative utterance and will therefore not be analysed in the present study (Maricchiolo, Gnisci, & Bonaiuto, 2012).

1.3.1 Gesture use in neurotypical individuals

Gesture use precedes the verbal communication of neurotypical individuals, but their occurrence decreases from the age of two and speech becomes the primary communication modality (Capone & McGregor, 2004; Stefanini, Bello, Caselli, Iverson, & Volterra, 2009). Nevertheless, gestures do not disappear and retain an important role in communication. Research mostly focusses on the support of substantial and beat gestures (Biau & Soto-Faraco, 2013; Clough & Duff, 2020; Igualada, Esteve-Gibert, & Prieto, 2017; Llanes-Coromina, Vilà-Giménez, Kushch, Borràs-Comes, & Prieto, 2018; Lucero, Zaharachuk, & Casasanto, 2014; McClave, 1994; Prieto et al., 2018). Both gesture types support the communication, but there is a shift in their presence during development. Around the age of five, neurotypical children use more substantial gestures than beat gestures, but only a few years later this balance reverses. The increase in use of beat gestures proceeds into adulthood (Colletta, Pellenq, & Guidetti, 2010; Florit-pons et al., 2020).

Substantial and beat gestures can support both the listener and the speaker. First, substantial gestures can aid listeners to comprehend the message during adverse listening conditions by showing the semantic information (Drijvers & Özyürek, 2017; Obermeier, Dolk, & Gunter, 2012), while beat gestures support speech understanding by accentuation of important words and parsing of the speech stream (Biau & Soto-Faraco, 2013; McClave, 1994; Prieto et al., 2018). Second, both substantial and beat gestures may aid the speakers with word retrieval (Igualada et al., 2017; Krauss, 1998; Krauss, Chen, & Gottesman, 2000). Additionally, substantial gestures support the speaker with transmitting spatial or motoric information (Alibali, 2005; Alibali, Heath, & Myers, 2001; Feyereisen & Havard, 1999; Hostetter & Alibali, 2007; Krauss, 1998; Rauscher, Krauss, & Chen, 1996) or with preparation and organisation of speech containing visuospatial information (Kita, 2000). Third, substantial and beat gestures create some cognitive benefits. Substantial gestures free-up working memory space (Cook, Yip, & Goldin-Meadow, 2012; Goldin-Meadow, Nusbaum, Kelly, & Wagner, 2001), they reflect the readiness of individuals to learn (Goldin-Meadow, Alibali, & Church, 1993) and information accompanied by substantial or beat gestures is better retained in memory (Cook, Duffy, & Fenn, 2013; Llanes-Coromina et al., 2018).

1.3.2 Gesture use in populations with speech and language impairment

While substantial and beat gestures can be used to support the communication of neurotypical individuals, the primary mode of communication in these individuals, remains speech (Capone & McGregor, 2004; Stefanini et al., 2009). In individuals with language and speech impairments, gestures appear to play a more important role during communication. In individuals with aphasia, for example, gestures can compensate for language problems and can sometimes become the primary communication mode (Hogrefe, Ziegler, Wiesmayer, Weidinger, & Goldenberg, 2013).

In individuals with dysarthria, where problems occur in the motoric movements of speech, both substantial and beat gestures appear to be able to increase the intelligibility of their speech. It was proposed that substantial gestures give supporting semantic information to the content of the speech, while beat gestures alter the prosody of the speech, which could make it more intelligible (Garcia & Cannito, 1996).

In individuals with Down syndrome, substantial gestures also appear to facilitate communication. In their early language development, they prefer using gestures or a bimodal form for communication over the verbal mode (Capone & McGregor, 2004; Chan & Iacono, 2001; Stefanini et al., 2009, 2007; Te Kaat-Van Den Os, Jongmans, Volman, & Lauteslager, 2015; Te Kaat-van den Os, Volman, Jongmans, & Lauteslager, 2017). Individuals with Down syndrome can use substantial gestures to compensate for their language problems (Lorang et al., 2018; Stefanini et al., 2007, 2008). Gesture use appears to be a strength in these individuals, therefore, they are often taught manual signs to support their verbal communication (Loncke, Nijs, & Smet, 2016). Research has shown that the stimulation of sign/gesture use early on in life in individuals with Down syndrome is beneficial for their language development (Chan & Iacono, 2001; Launonen, 2019; Layton & Savino, 1990; Te Kaat-Van Den Os et al., 2015) and their communication (Clibbens & Powell, 1994). For example, signs used during the pronunciation of the most important words of their messages appear to positively affect their speech intelligibility, even if the speaker cannot be seen by the listener (Clibbens & Powell, 1994).

1.4 The relation between gesture and speech/language

The research about individuals with speech and language impairments shows that these individuals use gestures in a compensating way. Herein, speech/language and gestures can be seen as two separate but interacting systems. However, there is also some evidence that gesture use might be affected by speech and language problems. Mayberry et al. (1998) examined gesture production in 12 neurotypical adults (22 - 58 years old) and four neurotypical children (11 years old) of whom half stuttered and half did not stutter. The gesture frequencies of the participants who stuttered were significantly lower than of those who did not stutter. Additionally, the participants who stuttered almost never co-produced a gesture with a stuttering event and the few gestures that did occur with a stuttering event fell to rest or froze mid-air until fluent speech resumed (Mayberry, Jaques, & DeDe, 1998). Mayberry et al. (1998) argued that the participants who stuttered made more extensive use of their available attentional resources of that system, which left fewer attentional resources for gesture production. This implies that speech is favoured as the first-priority mode of communication, so that gesture can only use the spare resources (Mayberry et al., 1998). Another study examined the effect of typical disfluencies on the gesture use of neurotypical children and of adults in their primary and secondary language. This study showed that gestures were almost always halted during typical disfluent moments (Graziano & Gullberg, 2018). These two studies seem to argue that speech/language and gestures belong to one integrated system.

Two other studies found a middle ground between these two contradicting views. Akhavan et al. (2016) also examined the relationship between gestures and typical disfluencies in neurotypical adults’ primary language. They discovered that the referential gestures often occurred during the disfluencies and that more disfluencies occur with a gesture than without a gesture. Nevertheless, they also found gestures that were disrupted by the disfluencies. Additionally, Rusiewicz et al. (2014) did a perturbation study wherein they altered the speech production of 15 American English speaking neurotypical participants to investigate the temporal alignment of gesture and speech. The participants viewed a screen with a question. There was a simultaneous spoken and gestural answer required. The answers were manipulated by presenting the participants with delayed auditory feedback (DAF). DAF leads to a disruption of the speech rhythm by altering the speech rate and increasing speech errors and disfluencies (e.g. Stuart et al., 2002; Yates, 1963). Prosodic stress was also manipulated by eliciting a contrastive pitch accent in the answers during the experiment. The results showed that the speech perturbations affected the gesture production, e.g. elongated speech corresponded with elongated total gesture time but did not reach significance.

Additionally, the coordination between gesture and vowel production did not remain coupled and revealed a loss of temporal alignment.

Both Akhavan et al. (2016) and Rusiewicz et al. (2014) found that gesture use could be affected by speech problems, however this was not continuous and often the gestures were unaffected or did not remain temporally aligned with the speech. In these cases, the gestures might even aid the communication. Therefore, they argued that gestures and speech/language are two closely interacting but still separate systems.

1.5 Purpose of the Study

Stuttering is a high-occurring speech problem in individuals with Down syndrome (Devenny & Silverman, 1990; Eggers & Van Eerdenbrugh, 2018; Kent & Vorperian, 2013; Van Borsel & Tetnowski, 2007). Individuals with Down syndrome spontaneously use gestures to communicate when their verbal communication is insufficient (Lorang et al., 2018; Stefanini et al., 2007) and they are often taught manual signs to improve their language development and facilitate their communication (e.g., Clibbens & Powell, 1994; Loncke et al., 2016). Therefore, it might be possible that these individuals are encouraged to use gestures or signs during stuttered speech. However, there is no research available about the impact of gestures on stuttering. Speech-language pathologists must understand the influence of one on the other before gesture use can be considered as a therapy option. Therefore, this exploratory study wants to have a first look at the interaction between gestures and stuttering in individuals with Down syndrome

The first aim is to investigate if gesture frequency differs between individuals with Down syndrome who stutter and who do not stutter, and to compare these results to neurotypical individuals. Present evidence points towards two potential influences. One the one hand, following the research of Mayberry et al. (1998), it is possible that stuttering leads to a lower gesture frequency in individuals with Down syndrome who stutter compared to the individuals with Down syndrome who do not stutter. Individuals with Down syndrome have intellectual disabilities and tasks require more cognitive effort from them compared to neurotypical individuals (Angulo-Chavira, García, & Arias-Trejo, 2017), which may result in an even lower gesture frequency according to Mayberry et al.'s (1998) argumentation.

On the other hand, if gesture and speech are controlled by two different motor systems that interact (Akhavan et al., 2016; Rusiewicz et al., 2014) and relieve cognitive load (Cook et al., 2013; Goldin-Meadow et al., 2001), then individuals with Down syndrome who stutter might be able to accompany the stuttering events with gestures as they do with their language problems (Lorang et al., 2018; Stefanini et al., 2007). In this case, gesture frequency would be higher in the individuals with Down syndrome who stutter. In the neurotypical individuals, we expect that the individuals who stutter will use fewer gestures than the neurotypical individuals who do not stutter as seen in Mayberry et al. (1998).

The second aim of the study is to investigate whether there is a difference in gesture frequency and gesture type (substantial and/or pragmatic) during stuttered versus fluent speech in individuals with Down syndrome and in neurotypical individuals. Language problems are compensated by substantial gestures in individuals with Down syndrome (Stefanini et al., 2007). Therefore, this study wanted to explore if stuttering events also elicit a specific type of gesture.

2. Material and method

2.1 Protocol

The following protocol was approved by The Ethics Committee Research of University Hospitals Leuven. The parents signed an informed consent and the participants also signed a consent form adjusted to their level of understanding. Parents were asked to film a spontaneous conversation with their child about their interests or daily activities. The participants needed to be visible from head to waist and without holding anything in their hands. The parents were asked to make several videos since stuttering is variable across topics (Arenas, 2017). On average, three videos were collected per participant. One video had a mean length of 2:31 minutes and the mean length of all the videos combined per participant was 8:25 minutes. Each participant had a conversation with his caregiver, who asked the participant questions to sustain a flowing conversation. The most occurring topics during the videos were school, trips, and hobbies. Additionally, the Peabody Picture Vocabulary test – III-NL (PPVT-III-NL; Dunn & Dunn, 2005), was administered. These test results were used to derive the developmental age of the participants with Down syndrome (Heath & Elliott, 1999; Klotzbier, Bühler, Holfelder, & Schott, 2020; Pueschel, Gallagher, Zartler, & Pezzullo, 1987; Reyes, Factor, & Scarpa, 2020). The developmental age of the neurotypical individuals is identical to their chronological age.

2.2 Participants

There were 31 participants with Down syndrome between the ages of 7 and 19 ($\overline{X}$ = 13 years 8 months) and 20 neurotypical participants between the ages of 3 and 10 ($\overline{X}$ = 6 years 2 months). All participants were able to understand and speak at least three-word sentences in Dutch. Parents were asked to fill out questionnaires about stuttering and additional problems. Parents of six of the 16 participants with Down syndrome that stutter (see 2.3 Groups) were uncertain or unaware of their child’s stuttering. In the neurotypical group, all the parents were aware of the stuttering of their children. None of the neurotypical children had additional problems, but all the individuals with Down syndrome had additional auditory, visual, motor, or intelligibility problems or had comorbid disorders (see Appendix A). Two individuals with Down syndrome were diagnosed with autism spectrum disorder. The presence of autism spectrum disorder has a significant impact on gesture use in individuals with Down syndrome (Lorang & Sterling, 2017) and therefore these two individuals’ data were excluded from analysis. The other problems did not lead to exclusion from the study since they were inherently associated with Down syndrome (Chen, 2006; Coppens-Hofman et al., 2012).

2.3 Groups

The participants with Down syndrome and neurotypical participants were each divided into a stuttering and non-stuttering group based on either their stuttering diagnosis by a stuttering therapist or their stuttering frequency and severity during spontaneous speech, calculated by the Test for Stutter severity_Non_Readers (TFS_NR; Boey, 2007). Table 1 shows the characteristics of each of these participant groups. The TFS_NR was calculated on the speech sample of the participant. One speech sample contained multiple videos of one participant. The goal, a speech sample of 300 words per participant, was not always possible due to their language skills. The speech samples had a mean of 391.57 words with a range from 100 – 955 words. The program CLAN (MacWhinney, 2000) was used to transcribe the speech samples of the participants and to code the stuttering events. Stuttering events were phoneme/syllable/monosyllabic repetitions, prolongations and blocks. The first author, who is a trained stuttering therapist, transcribed and coded the speech samples with assistance from Master of Science in Speech-Language Pathology students. In case an utterance was unintelligible to the transcribers, parents were contacted to aid in the transcription. Any remaining unintelligible words were coded with an X but still added to the total word count.

The TFS_NR takes secondary behaviours, type and length of stuttering events and the stuttering frequency into account. To calculate the stuttering frequency per participant, the stuttering events per 100 words were calculated ((total stuttering events/total words) x100) to account for the different lengths of the speech samples. Individuals with a minimum of 3% stuttering frequency (Boey, Wuyts, Van de Heyning, De Bodt, & Heylen, 2007) or a stuttering frequency between 2% and 3%, a TFS_NR score of at least 12 (corresponding with percentile 21) or a previous diagnosis by a stuttering therapist were placed in the stuttering group. In the

Down syndrome group, 16 participants stuttered and 13 were fluent. In the neurotypical group, 8 stuttered and 12 were fluent.

2.4 Controlled Measures

Besides the developmental age, we also measured stuttering frequency and severity, the chronological age, and Mean Length of the 5 Longest Utterances (ML5LU). The ML5LU gives an indication of the participants' language abilities. Language abilities can have an impact on the use of gestures. Younger children make proportionally more gestures than older children and the ratio of pragmatic and substantial gestures also changes with developmental age (Colletta et al., 2010; Florit-pons et al., 2020). Additionally, language problems might be compensated by gestures (Stefanini et al., 2007). Therefore, this variable was controlled for. The ML5LU was preferred over the Mean Length Of Utterance (MLU) since the latter is often lower due to the high occurrence of short utterances (Davis, 1937; Klee & Fitzgerald, 1985; Smith & Jackins, 2014). The ML5LU is therefore better suited to assess the expressive language ability of the participants (Smith & Jackins, 2014) and has been used in standardized language assessments, e.g. *The Renfew Bus Story Test* (Jansonius et al., 2014). To determine the ML5LU, the utterances were divided into communicative units or C-units following the rules provided in the SALT manual (Miller, Gillon, & Westerveld, 2012; Westerveld & Vidler, 2015). A C-unit was defined as either an independent clause with all its dependent clauses, an elliptical phrase, or as a sentence fragment defined as one complete thought based on the intonation pattern. Typical disfluencies were omitted from the word count of the ML5LU.

2.5 Dependent Measure

CLAN was also used to code the spontaneous hand gestures. Gestures were defined based on their gesture strokes (Bressem & Ladewig, 2011; Kendon, 1980). See Appendix B for an overview of the rules. Gestures were first coded according to their co-occurrence with or without speech. While traditionally co-speech gestures are accompanied by speech, sometimes an exception occurred, for example, if a child could not immediately retrieve the lexical item but could enact how to use it. Secondly, the function of the gestures was coded. The function of the gesture could be substantial, pragmatic (beat or other) or both. In case of doubt, a gesture was first defined as 'undefined' and given a category after deliberation. Three gestures remained undefined due to an unfavourable camera position. Some gestures of the individuals with Down syndrome were Key Word Signing (KWS) –signs (Loncke et al., 2016). The difference between spontaneous gestures and KWS signs is that the latter are learned signs with fixed parameters, while gestures occur spontaneously and have no predetermined form. These KWS signs only occurred in six participants and only one of them used them regularly. Omitting the signs from analyses would give distorted results since these signs could still be used to accompany stuttering events. Therefore, it was decided to code them as spontaneous substantial gestures because KWS signs have the same function as iconic gestures. Gesture frequency was measured as the number of gestures per 100 words (i.e., total gestures/total words x 100) to account for the different lengths of the speech samples (Wray, Saunders, McGuire, Cousins, & Norbury, 2017). Fluent and stuttered speech were also separately examined. Stuttered speech was defined as the total of stuttering events in the speech sample and fluent speech was the total of words minus the stuttering events. The gesture frequency during fluent speech was calculated as the number of gestures during 100 fluent words (i.e., total gestures during fluent speech/total fluent words x 100). The gesture frequency during stuttered speech was calculated as the number of gestures during 100 stuttering events (i.e., total gestures during stuttered speech/total stuttered speech x 100). Gesture type during fluent/stuttered speech (substantial, beat, other or both) are analysed as a proportion to the total amount of gestures during fluent/stuttered speech.

2.6 Statistics

2.6.1 Data analysis

To assess differences in background variables between the different groups, we compared ML5LU, chronological age, developmental age, stuttering frequency and severity for all the different comparison groups. Normally distributed variables were analysed with independent t-tests and non-normal distributed variables were analysed with Mann Whitney *U* tests. ML5LU and chronological age were normally distributed within each participant group according to the Shapiro-Wilk test and visual analyses, developmental age was normal in all the different comparison groups except for the entire Down syndrome and neurotypical group and the variables stuttering frequency and severity had a non-normal distribution. (Table 2).

Because multiple group comparisons were conducted per variable, a Holm-Bonferroni method was used to control the family-wise error rate. Table 2 and 3 showed that chronological age, ML5LU, stuttering frequency and severity differed significantly between the different comparison groups, while developmental age was not significantly different between the groups.

To analyse the first aim and examine if gesture frequency varies because of stuttering inside the Down syndrome and inside the neurotypical group, we used a multiple hierarchical linear regression. Because chronological age and ML5LU differed significantly between all the groups, they were entered as covariates in the first stage of the hierarchical regression. The group divisions of Down syndrome/neurotypical and stuttering/non-stuttering and their interaction effect were entered as binary predicting variables in the second stage of the hierarchical model. The tolerance scores and VIF scores of the multiple regressions were respectively above .2 and below .5, which indicates that there was no multicollinearity (Field, 2009).

To analyse the second aim and examine if gesture frequency and type differed between fluent and stuttered speech inside the Down syndrome and neurotypical group, we used a Wilcoxon signed-rank test because the differences in the gesture frequencies and in the different types of gestures between fluent and stuttered speech were not normally distributed (Field, 2009). The gesture frequency analyses during stuttered and fluent speech were chosen because some individuals with Down syndrome who do not stutter did have a few stuttering events but without belonging to the Down syndrome stuttering group, because they did not reach the 3% criterion. However, these stuttering events could also have an impact on the gesture production and complicate the comparison between the individuals who do and who do not stutter inside the Down syndrome group in the first aim of the study.

2.6.2 Reliability

To assess the reliability of the stuttering events and gesturing coding, researchers independently coded half of the data and measured interrater percent agreement. Two students trained in the coding systems recoded half of the speech of the individuals with Down syndrome, and one student recoded half of the speech of the neurotypical individuals. Interrater reliability for the stuttering events was 73.29% and 81.39% for respectively the Down syndrome and the neurotypical group. Interrater reliability for gesture coding was 87.88% and 79.46% for respectively the Down syndrome and the neurotypical group.

3. Results

Our first aim was to assess if gesture frequency differed between the different groups. We used a hierarchical multiple regression model. The model explains 33.0% of the variance in the gesture frequency. The covariates ML5LU and chronological age accounted for 24.3% of the variance and the groups accounted for the remainder 8.6%. While both significant, the first model was a better fit of the overall data, $F(2,48) = 7.397$, $p = .002$, than the second model, $F(5,48) = 4.299$, $p = .003$.

The coefficients in Table 4 show that individuals with Down syndrome had a significantly higher gesture frequency than neurotypical individuals. Between individuals who stutter and those who do not, no significant difference in gesture frequency was seen. There was no significant effect of the interaction between the presence of Down syndrome and stuttering on the gesture frequency. The variable ML5LU had a significant effect in the first stage but this disappeared when the predictor variables were entered.

In our second aim, we compared gesture frequency and type during fluent and stuttered speech inside the Down syndrome and neurotypical group. The participants with Down syndrome accompanied on average 12.64% of their fluent speech with gestures and 29.96% of their stuttered speech, while the neurotypical participants accompanied on average 4.29% of their fluent speech with gestures and 4.18% of their stuttered speech.

First, the Wilcoxon Signed Rank tests showed that the individuals with Down syndrome had a significantly higher gesture frequency during stuttered speech (Mdn = 23.08%) than during fluent speech (*Mdn* = 11.00 %), $T = -2.778$, $p = .004$, $r = -.52$. The neurotypical group did not show a significant difference in gesture frequency during fluent (Mdn = 3.92) and stuttered speech (*Mdn* = .00), $T = -1.531$, $p = .133$, $r = -.34$.

Second, the type of gestures during stuttered and fluent speech for both groups were analysed. Table 5 shows the absolute number and proportion of the different types during fluent and stuttered speech. The Wilcoxon Signed Rank test showed that there was no significant difference in frequency of a gesture type between fluent and stuttered speech in the individuals with Down syndrome (Table 6). The neurotypical individuals showed a significantly higher prevalence of substantial gestures, beat gestures and gestures with both functions during fluent speech than during stuttered speech (Table 6).

4. Discussion

The present study aimed to get a first insight into the interaction between stuttering and gesturing in individuals with Down syndrome. The gesture frequency of individuals with Down syndrome who stutter was compared to the gesture frequency of individuals with Down syndrome who do not stutter. These results were compared with those of neurotypical children. Additionally, we looked at the gesture frequency and gesture type during stuttered/fluent speech inside the Down syndrome and neurotypical group.

4.1 First aim

Our first aim was to see if individuals with Down syndrome who stutter and who do not stutter significantly differ in their gesture frequency. An hierarchical regression showed that there was a main effect of the presence of Down syndrome on the gesture frequency, but no main effect of the presence of stuttering and no interaction effect of Down syndrome/ neurotypical and stuttering/non-stuttering.

The significant difference in gesture frequency between the Down syndrome and neurotypical group appears to be in accordance with the research that shows that individuals with Down syndrome prefer bimodal communication (Lorang et al., 2018; Stefanini et al., 2007; Te Kaat-Van Den Os et al., 2015), while the neurotypical individuals prefer unimodal communication through speech (Capone & McGregor, 2004; Stefanini et al., 2009). However, the non-significance of a stuttering effect and of an interaction effect shows that gesture frequency was not affected by the presence of stuttering and that there was no difference in gesture frequency between the participants who stutter and who do not stutter inside the Down syndrome group or inside the neurotypical group, or between the two stuttering and the two non-stuttering groups. This result is unexpected considering the interaction of speech/language and gestures, either as an integrated system (Graziano & Gullberg, 2018; Mayberry et al., 1998) or as two separate systems (Akhavan et al., 2016; Rusiewicz et al., 2014). However, there could be some explanation for the current findings.

First, it could be that no interaction effect was detected because of the limited sample size of the groups. Second, while the individuals with Down syndrome who stutter and who do not stutter did differ significantly in their stuttering frequency and severity, there were still several stuttering events present in the individuals with Down syndrome who do not stutter, which might have influenced gesture use. Nevertheless, the non-significant difference between the stuttering and non-stuttering individuals from the neurotypical group shows that stuttering had no impact on their gesture frequency, and the difference cannot be accounted to other speech or language problems in this group. Fourth, while chronological age and ML5LU were controlled for, other communication aspects such as the topic of the conversation, might have influenced the use of gestures in the individual groups. If a participant is telling a story with a lot of spatial information, then the gesture frequency might be higher compared to someone who tells a story without spatial information, irrespective of their stuttering frequency (Alibali, 2005; Alibali et al., 2001; Feyereisen & Havard, 1999; Hostetter & Alibali, 2007; Krauss, 1998; Rauscher et al., 1996). The hierarchical regression showed that the model explained only 33.0% of the variance in the gesture frequency. Hereof only 8.6% was explained by the variables Down syndrome/neurotypical, stuttering/non-stuttering and their interaction effect, of which the latter two were non-significant. This confirms that there is still a lot of variance in gesture frequency that is unexplained.

Nevertheless, there appears to be some evidence that stuttering does not affect gesture use in neurotypical individuals. The results of the participants with Down syndrome are harder to interpret because of the stuttering events in the group of individuals with Down syndrome who do not stutter. Therefore, we also looked at the difference between stuttered and fluent speech inside the Down syndrome and neurotypical group using a pairwise model.

4.2 Second aim

For our second aim, we compared the gesture frequency and gesture type during stuttered speech and during fluent speech inside the Down syndrome and neurotypical group. The results of the Down syndrome group will be discussed first, followed by the results of the neurotypical group.

4.2.1 The Down syndrome group

The individuals with Down syndrome used significantly more gestures during stuttered speech than during fluent speech. This might be preliminary evidence that the individuals with Down syndrome have a spontaneous drive to compensate for their stuttering. This might mean that gesture use could be used in stuttering therapy considering Bray‘s (2015) suggestion to enhance and adjust the present behaviour.

However, there are some caveats that need to be resolved. First, the present study did not code if a gesture could (not) continue during the stuttering event. If the gestures were held as seen in Graziano and Gullberg (2018) and Mayberry et al. (1998) during the stuttering moments, then encouraging gesture use might lead to more interrupted gestures and to extra frustration. It is unknown if these gestures aided in the production and/or perception of the message. Second, while Bray’s theory on working with present behaviour might have benefits, no further research has been provided on how to administer this technique. Depending on the self-awareness of the individuals with Down syndrome, a focus on gesture use during stuttering might enhance negative feelings about their speech and have a negative impact on their self-esteem (Jackson et al., 2014). However, if they are not aware of their stuttering and the use of gestures is encouraged during all speech production, then this might be beneficial. Third, on average only 29.96% of their stuttered speech was accompanied by a gesture, so not all stuttering events triggered the occurrence of a gesture. This is in contradiction with Akhavan et al. (2016), who found more disfluent moments with a gesture than without a gesture. However, Akhavan et al. (2016) examined typical disfluencies while this study examined stuttering events, which might have a different influence on gesture use. The individuals with Down syndrome did not try to accompany every stuttering event with a gesture, but did use more gestures during stuttered speech than during fluent speech. It appears that they try to use gestures as a compensation strategy, but not consistently. It might be interesting to know what triggers gesture use: 1) the type of stuttering moment (repetition, prolongation, and block), 2) the ability of the individuals with Down syndrome to anticipate the stuttering moment or 3) the function of the word that the stuttering moment occurred on, e.g. only function words are supported by a gesture.

The type of gestures used during stuttered and fluent speech did not differ significantly. Substantial and beat gestures occurred at approximately the same frequency during stuttered speech (Table 5). This might mean that both gesture types can function as a compensatory gesture for stuttering. However, again, there was no analysis of the temporal coordination. So, either the substantial and beat gestures continued during the stuttering events and might have clarified the semantic meaning or affected the motoric production of the speech as suggested by Garcia and Cannito (1996) or the gestures were affected by the stuttering and could actually add to the stuttering severity.

4.2.2 The neurotypical group

There was no significant difference between gesture use during stuttered and fluent speech in the neurotypical participants, which is contrary to Mayberry et al. (1998). While no indication can be given about the temporal coordination between the gestures and the speech, stuttering did not appear to lead to a lower gesture frequency compared to fluent speech, nor did it lead to a higher gesture frequency as seen in the Down syndrome group. This might mean that the interaction between gestures and stuttering is different in neurotypical individuals compared to individuals with Down syndrome. However, this research should be repeated, including neurotypical individuals with a higher stuttering severity. In the group of neurotypical individuals who stutter, the stuttering frequency remained below 6%, while that of the group of individuals with Down syndrome who stutter reached above 20%. The frequency of stuttering events might have been too low in the neurotypical group to see if there was a difference in gesture frequency between fluent and stuttered speech.

The neurotypical individuals used more substantial gestures, beat gestures and gestures with both functions during fluent than during stuttered speech. This shows that none of the gesture types was inclined to occur more during stuttering events than during fluent speech. However, the low number of stuttering events accompanied by a gesture in the neurotypical group (15) might make interpretation of these results difficult.

4.3 Limitations and Future Research

Individuals with Down syndrome use more gestures during stuttered speech than during fluent speech, which might indicate that they try to use gestures to compensate for stuttering events. However, the current study is only preliminary and we should be cautious with the above interpretations due to the small group sizes. Future research should include more participants.

Some other limitations should also be taken into account. First, the developmental age of the participants was calculated from the PPVT-III-NL as done in earlier studies (Heath & Elliott, 1999; Klotzbier et al., 2020; Pueschel et al., 1987; Reyes et al., 2020). However, it might have been too restrictive to base their developmental age on one test for vocabulary. The developmental age of the neurotypical individuals was assumed to be equal to their chronological age, as they had no additional language or speech problems. Nevertheless, there could still be some deviations from the norm. In future research, developmental age should be based on a broader test including both verbal and non-verbal skills and should be administered to both the Down syndrome and the neurotypical participants.

Second, language proficiency was significantly different between the stuttering and non-stuttering group of the individuals with Down syndrome according to the ML5LU. While we did account for this in the analyses, this is still a limitation. A lower ML5LU may indicate language difficulties and, therefore, a higher gesture use can be expected in individuals with lower ML5LU (Stefanini et al., 2007). The non-significant difference in gesture frequency between the stuttering and non-stuttering group of the individuals with Down syndrome could be caused by the imbalance between speech and language problems. If gestures are used to compensate for both, then the individuals with Down syndrome who stutter probably used them primarily in the context of stuttering, as their mean ML5LU was quite high, and the individuals with Down syndrome who do not stutter in the context of language production because of their lower ML5LU. Future research should therefore try to match individuals on language production level.

Third, language may indeed play a role. The present study only looked at the stuttering events to investigate the gesture-speech relationship, because Mayberry et al. (1998) found that normal disfluencies did not affect gesture use. However, more recent research found a relationship between gestures and normal disfluencies (Akhavan et al., 2016; Graziano & Gullberg, 2018). Therefore, it would be interesting to compare gesture use during normal disfluencies and stuttered speech in future research. Hereby, it would be interesting to analyse the temporal relationship at word level to see if the gesture stopped or continued during the stuttering event/typical disfluency. It would also be interesting to investigate the interaction between KWS-signs and stuttering at the word-level. It was presumed that excluding the KWS from the analysis would give distorted results, as they might be used to accompany stuttering events. However, KWS-signs are taught to the individuals to facilitate their communication (Loncke et al., 2016) and might have been produced primarily to support the meaning of the word irrespective of the stuttering event. Some individuals with Down syndrome use KWS as their primary form of communication and therefore almost all their stuttering events would be accompanied by a sign, without the specific function of coping with the stuttering event. While only one participant used KWS-signs regularly, future research should aim to accommodate for these signs. It should be investigated if KWS-signs can facilitate the communication of utterances with stuttering events and if their relationship with stuttering is different from the relationship between spontaneous gestures and stuttering.

Clibbens and Powell (1994) already showed that KWS-signs could increase the intelligibility of speech of individuals with Down syndrome. It might also be interesting to look if KWS-signs and gestures can affect the intelligibility of stuttered events. If gestures/KWS-signs continue during stuttering events and bolster speech intelligibility, it might be beneficial to promote gesture use/KWS-signs in individuals with Down syndrome who stutter.

5. Conclusions

The present research explored the speech-gesture relationship in individuals with Down syndrome and compared this with neurotypical individuals. The results showed a significant difference in gesture frequency between the Down syndrome and neurotypical group. This significant difference may support the idea that individuals with Down syndrome are more inclined to communicate bimodal than neurotypical individuals. However, this difference does not give information about the speech-gesture relationship as both groups included individuals who stutter and who do not stutter. There appeared to be no significant difference in gesture use between any of the other comparison groups, which may suggest that stuttering does not affect gesture use. However, when analysing gesture use inside the Down syndrome and the neurotypical group, there was some evidence for an attempt to compensate stuttered speech in the Down syndrome group. Nevertheless, the majority of the stuttering events occurred without a gesture. This implies that gestures might be used to compensate for stuttering events, but it remains unknown what triggers the compensation reaction and whether the compensation is successful. Further research is necessary into the speech-gesture relationship of individuals with Down syndrome. If individuals with Down syndrome are able to accompany stuttering events with gestures and improve their fluency and comprehensibility, then gesture use should be investigated as a therapy method for individuals with Down syndrome who stutter.

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7. Data availability statement

The data that support the findings of this study are available from the corresponding author, [BM], upon reasonable request.

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Table 1

Characteristics of the different participant groups

	CL_age (y;m)	DA_age (y;m)	ML5LU	Stuttering frequency (%)	Stuttering severity (Pc)	Gesture frequency (%)
DS	13;8 (6;2 – 19;9)	5;4 (2;10 – 11;12)	9.29 (4.00-15.60)	5.74 (.00- 22.99)	26.72 (0 – 90)	14.04 (.00 – 39.00)
NT	6;5 (3;3 – 10;5)	6;5 (3;3 – 10;5)	13.14 (9.40 - 18.40)	1.53 (.00-5.07)	8.90 (0 - 35)	4.71 (.15 -8.06)
DSWS	15;2 (8;0 – 19;9)	5;8 (2;10 – 11;12)	10.33 (7.40 – 15.60)	9.61 (2.41 – 22.99)	44.44 (16 – 90)	11.58 (.00 – 25.65)
DSNS	8;9 (6;2 – 17;11)	4;11 (2;9 – 9;6)	8.02 (4.0 – 11.40)	0.98 (.00 – 2.4)	4.92 (0 - 13)	17.07 (.78 – 39)
NTWS	5;8 (3;10 – 9;11)	5;8 (3;10 – 9;11)	12.93 (9.40 - 18.40)	3.77 (.91 – 5.07)	21.75 (12 - 35)	4.35 (.60 – 7.78)
NTNS	6;3 (3;3 – 10;5)	6;3 (3;3 – 10;5)	13.28 (10.00 – 18.20)	.05 (.00 - .30)	0.33 (0 - 3)	4.94 (.15 - 8.06)

Note. DS= Individuals with Down syndrome, NT = Neurotypical individuals, DSWS = Individuals with Down syndrome who stutter, DSNS = Individuals with Down syndrome who do not stutter, NTWS = Neurotypical individuals who stutter, NTNS = Neurotypical individuals who do not stutter, CL_age = Chronological age, DA_age = Developmental age, ML5LU is the Mean Length of the 5 Longest utterances. The cells display the mean and range of the different variables within each group.

Table 2

Comparisons of Developmental Age, Chronological Age and ML5LU between the participant groups

Group Comparison		*Mean*	*p*	*r*	*95% CI*
	Developmental age (y;m)†	5;4 – 6;5	.226	.17	[-.117; .431]
DS - NT	Chronological age (y;m)	13;8 – 6;5	< .001*	.79	[.654;.876]
	ML5LU (w/u)	9.29 – 13.14	<.001*	.61	[.397; .761]
	Developmental age (y;m)	5;8 - 4;11	.377	.17	[-.210; .505]
DSWS - DSNS	Chronological age (y;m)	15;2 - 8;9	.016*	.44	[.088; .695]
	ML5LU (w/u)	10.33 - 8.02	.008*	.48	[.138; .720]
	Developmental age (y;m)	5;8 - 6;3	.572	.13	[-.332; .541]
NTWS - NTNS	Chronological age (y;m)	5;8 - 6;3	.572	.13	[-.332;.541]
	ML5LU (w/u)	12.93 – 13.28	.777	.07	[-.384; .497]
	Developmental age (y;m)	5;8 – 5;8	.992	.00	[-.402; .405]
DSWS - NTWS	Chronological age (y;m)	15;2 – 5;8	<.001*	.83	[.641; .924]
	ML5LU (w/u)	10.33 – 12.93	.021*	.47	[.082; .734]
	Developmental age (y;m)	4;11 - 6;3	.138	.31	[-.097; .628]
DSNS - NTNS	Chronological age (y;m)	8;9 - 6;3	<.001*	.67	[.374; .842]
	ML5LU (w/u)	8.02 – 13.28	<.001*	.76	[.521; .888]

Note. (y;m) = years; months, DS= Individuals with Down syndrome, NT = Neurotypical individuals, DSWS = Individuals with Down syndrome who stutter, DSNS = Individuals with Down syndrome who do not stutter, NTWS = Neurotypical individuals who stutter, NTNS = Neurotypical individuals who do not stutter. ML5LU is the Mean Length of the 5 Longest Utterances in a participant's speech sample.

† Developmental age was compared with a non-parametric Mann Whitney U test because of a non-normal distribution of developmental age in the entire Down syndrome and neurotypical population.

*Significant at $p \leq 0.05/(5 –$ rank number of pair (by degree of significance) + 1), Holm-Bonferroni Method.

Table 3

Comparisons of Stuttering Frequency (SF) and Stuttering Severity (SS)

Group Comparison	Measure	Median †, ‡	*p*	*r*	*95% CI*
DS - NT	SF	3.06% - .13%	.002*	-.44	[-.642; -.181]
	SS	Pc 21 – Pc .50	.004*	-.40	[-.612; -.134]
DSWS - DSNS	SF	5.69% - 0.78%	<.001*	-.85	[-.928;-.702]
	SS	Pc 32.50 – Pc 5	<.001*	-.85	[-.928; -.702]
NTWS - NTNS	SF	4.26% - .00%	<.001*	-.89	[-.956; -.738]
	SS	Pc 19.50 – Pc 0	<.001*	-.89	[-.956; -738]
DSWS - NTWS	SF	5.69% - 4.26%	.038*	-.42	[-.704; -.020]
	SS	Pc 32.50 – Pc 19.50	.018*	-.48	[-740; -.095]
DSNS - NTNS	SF	0.78% - .00%	<.001*	-.73	[-.873; -.471]
	SS	Pc 5 – Pc 0	.001*	-.63	[-.821; -.313]

Note. DS = Individuals with Down syndrome, NT = Neurotypical individuals, DSWS = Individuals with Down syndrome who stutter, DSNS = Individuals with Down syndrome who do not stutter, NTWS = Neurotypical individuals who stutter, NTNS = Neurotypical individuals who do not stutter. † The first value represents the median of the first group in the 'group comparison' column and the second value the median of the second group. Median is preferred over mean in non-parametric tests (Field, 2009). ‡A higher Pc score reflects a higher stuttering severity.

*Significant at p ≤ 0.05/(5 – rank number of pair (by degree of significance) + 1), Holm-Bonferroni Method

Table 4

Coefficients, p-value, 95% confidence interval and effect size for comparison of gesture frequency at the first and second stage of the hierarchical linear multiple regression.

Variable	*β*	*t*	*p*	*95.0% Confidence interval*	*Effect size*
Stage 1					
ML5LU	-1.302	-3.339	.002**	[-2.086; -.0517]	-.442
Chronological age	.021	1.049	.300	[-.020; .062]	.153
Stage 2					
ML5LU	-.465	-.886	.381	-1.524; .594	-.134
Chronological age	-.021	-.629	.532	-.090; .047	-.096
DS versus NT	11.072	2.139	.038*	.635; .21.508	.310
Stuttering versus non-stuttering	-.905	-.247	.806	-8.300; 6.490	-.038
Interaction effect	-2.622	-.519	.606	-12.809; 7.566	-.079

Note. DS = Individuals with Down syndrome, NT = Neurotypical individuals

*p < .05. **p < .01. ***p < .001

Table 5

Descriptive data on the gesture types that accompanied stuttered/fluent speech

	Group	Total *Ges* *%*	Substantial *Ges* *%*	Beats *Ges* *%*	Other *Ges* *%*	Both *Ges* *%*	Undefined *Ges* *%*
During stuttered speech	DS	138 100	62 44.93	64 46.38	7 5.07	5 3.62	/
	NT	15 100	11 73.33	3 20	1 6.67	0 0	/
During fluent speech	DS	1324 100	690 52.11	464 35.05	54 4.08	113 8.53	3 .23
	NT	364 100	177 48.63	131 35.99	31 8.52	25 6.87	/

Note. DS = Individuals with Down syndrome, NT = Neurotypical individuals. Ges = The number of gestures of that type that accompanied stuttered/fluent speech. % = Percentage of gestures of that type that accompanied stuttered/fluent speech. The percentage is calculated by $\frac{Number\ of\ (type) gestures\ with\ stuttered/fluent\ \ speech}{Number\ of\ gestures\ with\ stuttered/fluent\ \ speech}$.

Table 6

Wilcoxon Signed Rank test for difference in use of gesture types between fluent and stuttered speech

	Measure	Gestures during stuttered speech *Median (%)*[a] *SD (%)*	Gestures during fluent speech *Median (%)*[a] *SD (%)*	Wilcoxon Signed Rank Test *T* *p-value* *Effect-size*
DS	Substantial	25.00 33.50	50.00 24.58	-2.403 .015
	Beats	22.22 33.52	39.02 21.23	-.991 .331
	Other pragmatic	0 7.48	0 3.94	-1.712 .094
	Both	0 15.22	0 15.73	-.889 .413
NT	Substantial	0 36.36	57.78 27.99	-3.321 <.001*
	Beats	0 22.75	23.21 20.34	-2.510 .010*
	Other pragmatic	0 2.80	5.4 7.50	-2.668 .005*
	Both	0 0	0 16.15	-2.023 .063

Note. DS = Individuals with Down syndrome, NT = Neurotypical individuals. The percentages of types are measured as the total of substantial gestures during fluent/stuttered speech divided by the total of gestures during fluent/stuttered speech. [a] Median is preferred over mean in non-parametric tests (Field, 2009)

* Significant at $p \leq 0.05/(4 – \text{rank number of pair (by degree of signifcance)} + 1)$, Holm-Bonferroni Method.

Appendix A

Table A1

Co-occurring problems in the participants with Down syndrome

Participant	ASD	Visual problems	Auditory problems	Motor problems	Other speech problems	Worst Intelligibility Score (1-7)	Best Intelligibility Score (1-7)
1		X	X	X		2	2
2		X	X	X	X	5	3
3		X	X		X	5	3
4		X				2	2
5						4	4
6						5	5
7						4	2
8						4	4
9						5	3
10		X		X		5	3
11				X		4	2
12	X					4	4
13						4	1
14						3	3
15						4	2
16		X				5	3
17		X	X	X		4	4
18	X	X				4	4
19		X				6	2
20		X	X	X		2	2
21		X			X	5	3
22						4	4
23				X		2	1
24				X	X	4	3
25		X		X		5	3
26				X		2	2
27		X	X		X	2	2
28		X			X	4	4

29		X			X	4	3
30		X		X	X	4	4
31						4	2
Total participants	2	16	6	11	8	3.9 (1.106)[ł]	2.8 (.991)[ł]

Note. Intelligibility can differ between situations, therefore parents were asked the rate the intelligibility of their child on a 7-point Likert scale developed by Strand and Skinder (1999) in a difficult and an easy speaking situation. The scores corresponded to the descriptions below. [ł]Mean and standard deviation of the intelligibility scores of the participants.

1 = Completely intelligible, no differences with conventional speech

2 = Intelligible, but sometimes differences with conventional speech

3 = Intelligible, but many differences with conventional speech.

4 = Intelligible with extra attention to the speech, with some unintelligible words.

5 = Difficult to understand, many unintelligible words

6 = Most of the time unintelligible

7 = Completely unintelligible

Appendix B

Defining a gesture

A gesture consists out of five phases: a rest position, a preparation phase, a stroke, a hold and a retraction phase (Bressem & Ladewig, 2011; Kendon, 1980). In the rest position, there is no movement of the hands. In the preparation phase, the hands move from rest position to prepare for the stroke. The stroke is the most important part of the gesture, since it depicts the intention of the gesture. This stroke is sometimes accompanied by a hold phase, a motionless phase right before or after the stroke. A gesture ends with a retraction phase, where the hands return to rest position after the stroke. However, during communication the rest position, preparation and/or the retraction phase are often missing, so that strokes (with or without hold) follow each other instantaneously.

Therefore, since a stroke phase is necessary to speak of a gesture, every new stroke phase (with or without the preceded and following phases) was counted as a new gesture. To determine a gesture when certain phases are missing, the following rules were applied:

1) Different gestures were defined if they were divided by a pause or an obvious change in shape or trajectory (Jacobs & Garnham, 2007; Kong, Law, Kwan, Lai, & Lam, 2015).
2) If a stroke was hold over multiple verbal utterances of one person, without a change in form, position or trajectory, then this was coded as one gesture.
3) Multiple strokes that depicted the same meaning were counted as different gestures. For example, if somebody said ‘She fell three stairs down’, while making three times the same downward movement with the palm of the hand orientated downwards on the words ‘three’, ‘stairs’ and ‘down’, then this was counted as three gestures. However, there was one exception. If the repeated movement was an integral part of

the action, such as scraping, patting and stirring, then these strokes were counted as one gesture (Müller et al., 2014).

4) If a stroke was interrupted by a stuttering moment, but continued afterwards, then this also counted as one gesture.