RUNNING TITLE: SPEECH DISFLUENCIES IN DOWN SYNDROME

Speech Disfluencies in Children with Down Syndrome

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Abstract

Purpose: Speech and language development in individuals with Down syndrome is often delayed and/or disordered and speech disfluencies appear to be more common. These disfluencies have been labeled over time as stuttering, cluttering or both. Findings were usually generated from studies with adults or a mixed age group, quite often using different methodologies, making it difficult to compare findings. Therefore, the purpose of this study was to analyze and describe the speech disfluencies of a group, only consisting of children with Down Syndrome between 3 and 13 years of age.

Method: Participants consisted of 26 Dutch-speaking children with DS. Spontaneous speech samples were collected and 50 utterances were analyzed for each child. Types of disfluencies were identified and classified into stuttering-like (SLD) and other disfluencies (OD). The criterion of three or more SLD per 100 syllables (cf. Ambrose & Yairi, 1999) was used to identify stuttering. Additional parameters such as mean articulation rate (MAR), ratio of disfluencies, and telescoping (cf. Coppens-Hofman, et al., 2013) were used to identify cluttering and to differentiate between stuttering and cluttering.

Results & Conclusion: Approximately 30 percent of children with DS between 3 and 13 years of age in this study stutter, which is much higher than the prevalence in normally developing children. Moreover, this study showed that the speech of children with DS has a different distribution of types of disfluencies than the speech of normally developing children. Although different cluttering-like characteristics were found in the speech of young children with DS, none of them could be identified as cluttering or cluttering-stuttering.

Key words: speech disfluencies, Down Syndrome, stuttering, cluttering

Introduction

Individuals with Down Syndrome (DS) have a complete or partial copy of chromosome 21 resulting in a typical appearance (Shprintzen, 1997), with features such as broad hands, flat nasal bridge, lax ligaments, intellectual disabilities, open mouth and short posture. Prevalence numbers are estimated around one in 732 to 1000 babies in the US (e.g., Roizen & Patterson, 2003; Sherman, Allen, Bean & Freeman, 2007) and around one in 450 to700 babies in Europe (Loane et al., 2013; Irving, Basu, Richmond, Burn, & Wren, 2008). DS tends to occur more frequently when mothers are 35 or older when pregnant. One could expect that the number of foetuses with DS is increasing as a consequence of the rising average age of maternity (e.g., Loane et al., 2013). The increased number of prenatal screening and termination of pregnancy, however, counterbalances this expected rise of prevalence estimates, and keeps these numbers relatively stable. Prevalence estimates however, tend to vary among racial and ethnic populations.

Children with DS usually receive support early in life thanks to its easy identification (Roizen & Patterson, 2003). Children with DS are encouraged to enter the education system at an early age to stimulate development and inclusion (with support) in the classroom. This early intervention approach recognizes the importance of being able to communicate with other children. Hence, it is important to understand the speech patterns of children with DS.

Children with DS are thought to acquire speech and language more slowly than children without DS (e.g., Chapman & Hesketh, 2001; Miller, 1992; Rondal, 1995), despite considerable individual variability. The acquisition of first words seems to be delayed (e.g., Berglund, Eriksson, & Johansson, 2001; Stoel-Gammon, 2001) and also the subsequent

growth of expressive vocabulary has been found to occur at a slower pace (Buckley & Rondal, 2003). Syntax, both receptive and expressive, seems to be more impaired than vocabulary (e.g., Caselli, Monaco, Trasciani, & Vicari, 2008; De Bal, 2005; Price et al., 2008) and also in the pragmatic aspects of language, weaknesses have been shown (e.g., Abbeduto & Hesketh, 1997). Van Borsel (1993) discussed both the presence of phonological and phonetic errors in the speech of children with DS and phoneme-level research indicates that between 5 and 10 years of age, children with DS do not have a simple delay in acquiring phonemes (Dodd & Thompson, 2001; Zisk & Bailer, 1967). Repeated word productions of children with DS contain more inconsistent errors than the repeated word productions of children without DS. Syllable structure phonological processes such as cluster reduction, final consonant deletion, use of stops for fricatives (Dodd & Thompson, 2001; Stoel-Gammon, 2001) seem to be common and especially the inconsistency of errors seems characteristic in the speech of children with DS (Dodd & Thompson, 2001). Based on a literature review on articulation and phonology studies conducted in children with DS, Kent and Vorperian (2013) concluded that a combination of delayed (i.e. developmental) and disordered (i.e., non-developmental) patterns were found. In combination with apraxia of speech and speech dysarthria (Martin, Klusek, Estigarribia, & Roberts, 2009), these speech patterns may lead to decreased speech intelligibility, a consistent finding in children with DS (Barnes et al., 2009; Kent & Vorperian, 2013; Kumin, 2001).

It is known that the speech of individuals with DS is often not fluent (Bray, 2003, 2008; Coppens-Hofman, et al., 2013; Devenny & Silverman, 1990; Preus, 1972; Van Borsel, 1993, 2011; Van Borsel, & Tetnowski, 2007; Van Borsel & Vandermeulen, 2008; Withaegels, 2009). The odds ratio of a fluency disorder in children between 3 and 17 years old with DS is 0.6 to 1.8 compared to individuals with an intellectual disability (Schieve,

Boulet, Boyle, Rasmussen, & Schendel, 2009), a group in which speech disfluencies are already more common than the normal population (Coppens-Hofman et al., 2013). Even though individuals with DS are more often presenting with a fluency disorder, it is not a typical feature of DS (Kent & Vorperian, 2013).

The estimated prevalence of stuttering in individuals with DS has been reported to range from 15% to 45% (Bloodstein, 1995; Devenny & Silverman, 1990; Van Riper, 1971). Some studies provided more detailed analyses of the types of disfluencies. Cabanas (1954) reported repetitions, blocks (mostly on vowels), hesitations and hurried speech. Willcox (1988) found sound, syllable, and word repetitions, phrase repetitions and also prolongations. In Bray's (2003) questionnaire-based research repetitions of sounds and syllables, interjections, blocks, and prolongation of sounds were among the most identified disfluencies. Although these reported types of disfluencies seem to be a combination of both stuttering-like (SLD) and other disfluencies (OD; Ambrose & Yairi, 1999), they have clearly identified core stuttering characteristics in the speech of individuals with DS since stuttering is defined in the International Classification of Diseases (World Health Organization, 1993) as

> speech that is characterized by frequent repetition or prolongation of sounds or syllables or words, or by frequent hesitations or pauses that disrupt the rhythmic flow of speech. There may be associated movements of the face and/or other parts of the body that coincide in time with the repetitions, prolongations, or pauses in speech flow. (p. 227)

With regard to the associated movements or secondary stuttering behaviors, findings are somewhat less unequivocal. While some authors reported secondary behaviors, i.e.,

struggling, such as eye blinking and facial grimaces, avoidance and postponement behaviors (Bray, 2008; Devenny & Silverman, 1990; Preus, 1972; Schlanger & Gottsleben, 1957; Stansfield, 1990), others did not find any secondary characteristics related to these disfluencies (Cabanas, 1954; Van Riper, 1971). The absence of secondary behaviors (Van Borsel & Tetnowski, 2007) together with only limited or no levels of awareness, anxiety or avoidance (Bray, 2008) led some authors (e.g. Cabanas, 1954; Otto & Yairi, 1975) to conclude that this disfluency pattern is more representative of cluttering than stuttering.

Myers, Bakker, St. Louis and Raphael (2012), define cluttering as "a fluency disorder characterized by various symptoms such as poor intelligibility, a speaking rate perceived to be too fast or irregular, inappropriate prosody, as well as the presence of disfluencies" (p. 9). Based on his speech analyses of 47 individuals with DS, Preus (1972) concluded that in 28% could be diagnosed as people who stuttered, 13% as people who cluttered, and 19% as people with combined stuttering and cluttering. Based on 76 questionnaires filled out by 26 speech language therapists (SLTs) working with individuals with DS, Van Borsel and Vandermeulen (2008) concluded that 79% could be classified as persons who clutter and 17% as persons with both cluttering and stuttering. Somewhat more recent, also Coppens-Hofman et al. (2013) analyzed the speech of 28 adults with intellectual disabilities, of which 11 were diagnosed with DS, and identified 46% as persons who clutter, 29% with combined stuttering and cluttering and 25% with no fluency disorder.

It is clear that there is some disagreement about how to classify the fluency disorder in the speech of individuals with DS. The methods used to identify and label the fluency disorder in the speech of individuals with DS varied considerably in the three studies

mentioned above. This may have had an impact on the prevalence estimates, which varied considerably. Van Borsel and Vandermeulen (2008) utilized the Predictive Cluttering Inventory (Daly & Cantrell, 2006). This inventory contains 50 signs and symptoms that characterize cluttering. SLTs completed the inventory for their clients. Preus (1972) included judges (i.e., individuals who knew the participants well) to rate the speech samples perceptually, using a 4-point scale to indicate the presence of cluttering and a 5-point scale for stuttering. Coppens-Hofman et al. (2013) attempted to use the standardized procedure suggested by van Zaalen-op ‘t Hof, Wijnen and De Jonckere (2009), albeit with the necessary adaptations. They looked into several parameters including Mean Articulation Rate (MAR), types of disfluencies and telescoping, a linguistic phenomenon referring to the merging of syllables, such as ‘horfic’ for ‘horrific’. Despite the different methods applied in the three studies, it became apparent that there is a need for more detailed investigation of the fluency disorder in people with DS.

It is not common to find a study that includes young children with DS, and even less common to find a study that investigated these young children as a separate group. Van Borsel and Vermeulen (2008) included children as young as 3 years of age, and found that results from this group (children younger than 10 years of age) deviated from the other age groups. That is, SLTs left many items on the Predictive Cluttering Inventory (Daly & Cantrell, 2006) unscored for this group of children. Children with DS as young as 3 years of age, for whom fluency disorders were investigated, were not yet included in any other study (Kent & Vorperian, 2013). Hence, the goal of the current study was to analyze and describe the different types of disfluencies of young children with DS.

Method

Participants

Participants were 26 monolingual Dutch-speaking children (12 boys and 14 girls) with DS, aged between 3;03 (40 months) and 12;06 years of age (151 months). The mean age was 8;07 years (SD = 2;10 years). All children (1) were diagnosed with DS by a medical team, (2) were able to speak and (3) were younger than 13 years of age. The children were recruited from several institutions and schools, and through parent committees of children with DS in Flanders, the Northern part of Belgium. The study was approved by the Lessius Research Council and ethical committees of the participating institutes. None of the participating children were specifically diagnosed with stuttering and/or cluttering previously, or received specific treatment for fluency disorders. Due to the educational system in Flanders, most children with DS receive speech-language treatment by SLTs working in the school or private system. Characteristics of the participants can be found in Table 1.

INSERT TABLE 1 ABOUT HERE

Collection of the data

Spontaneous speech samples were audio-recorded during a 15-minute play session with a toy farm and/or a colored picture book with different scenes. Depending on the age and the interests of the child, more emphasis was placed on the first or the latter. These conversations took place in a separate room in the school or institution where the child resided, or in their home environment. The speech samples were collected several years ago with a Panasonic tape recorder, type RQ-L11, as part of a series of studies (Eggers, 2010; Eggers & De Bal, 2009) looking into the disfluent speech patterns in various population subgroups. Subsequently, 50 consecutive utterances were selected from the original

recordings and digitalized to perform the analysis. According to Darley and Moll (1960), a speech sample of 50 utterances has a reliability coefficient of 0.85 and is "adequate for most purposes" (p. 128) (see also Craig, Washington, & Thompson-Porter, 1998; Hutchins, Brannick, Bryant, & Silliman, 2005; Zackheim & Conture, 2003). Similar to Johnson, Darley, and Spriestersbach (1963) an utterance was defined as each individual thought, separated from the previous and followed by a pause. No utterances were selected out of the first two minutes of the sample in order to make sure the child was acquainted with the situation. Utterances that were unintelligible, or that consisted of simple yes-no answers or onomatopoeias were also deleted. Number of words and syllables for each participant are given in Table 1, as well as average number of syllables per utterance and mean length of utterance (MLU) in words. Since mean length of utterance (MLU) based on words is almost perfectly correlated with MLU based on morphemes and can be used as effectively in the measurement of the child's gross language development (Parker & Brorson, 2005), MLU was calculated based on words. Not all speech samples contained 50 usable utterances; hence, the maximum possible utterances were selected.

Categorization of disfluencies

The selected utterances were loaded into and transcribed in the PRAAT software (Boersma & Weenink, 2005), which enables accurate speech analysis. Based on both visual and auditory inspection, disfluencies were identified and categorized. Even though categorizing disfluencies may be questioned to be a reliable measure to identify stuttered speech in the daily clinical practice (Einarsdóttir & Ingham, 2005), it is an essential component in studies that attempt to describe the disfluent speech, such as this study. Despite the controversy that using categories can elicit, it is a widely used method to describe

disfluent speech (e.g., Anderson, Pellowski, Conture & Kelly, 2003; Coppens-Hofman et al., 2013; Yairi & Ambrose, 1992; 1999; Yairi, Ambrose, Paden & Throneburg, 1996).

INSERT TABLE 2 ABOUT HERE

Existing classification systems for disfluencies are usually based on Johnson's initial classification system (Johnson et al., 1959; Johnson, 1961), albeit adapted over the years in different studies. The classification system used in this study (Table 2) is also based on Johnson's original work, but was elaborated from eight categories (original classification) to nine categories, similar to what Yairi and Ambrose (1999) did. Certain categories are commonly used to define 'stuttering-like disfluencies' (SLD; Yairi & Ambrose, 1992), such as blocks and prolongations (e.g., Ambrose & Yairi, 1999; Coppens-Hofman et al., 2013; Guitar, 2006; Tumanova, Conture, Lambert, & Walden, 2014; Yairi & Ambrose, 1999; Zackheim & Conture, 2003). Similarly, certain categories such as interjections and revisions are commonly used to define 'non-stuttered disfluencies', 'other disfluencies' (OD) or 'normal disfluencies' (e.g., Ambrose & Yairi, 1999; Coppens-Hofman et al., 2013; Tumanova et al., 2014). In this study, sound and syllable repetitions (i.e., part-word repetitions and single syllable word repetitions), prolongations, blocks, and broken words were labeled SLD, similar to the Illinois studies, where the latter three categories were clustered under 'disrythmic phonations' (Yairi & Ambrose, 1992; 1999; Yairi et al., 1996). Phrase repetitions, multisyllable word repetitions, interjections, and revisions were labeled OD.

Transcription and identification of the disfluencies was done by the second author, after two joint training sessions with the first author. The second and first author has respectively over 10 and 15 years of experience in this kind of data analysis. The first author independently labeled and categorized 20% of the utterances and interjudge reliability (point-by-point for location and type, see Ambrose & Yairi, 1999) was calculated based on the 'agreement index' percentage, i.e. amount of agreements divided by the sum of agreements and disagreements (Suen & Ary, 1989). The interjudge reliability was .91.

Diagnostic criterion for stuttering

The generally accepted criterion to decide whether a child stutters is the presence of three SLD or more per 100 syllables (e.g., Ambrose & Yairi, 1999; Bloodstein, 1995; Yairi & Ambrose, 1992, 1996; Yairi et al., 1996; Zackheim & Conture, 2003). A large study with Dutch-speaking preschool age children confirmed that the 3% criterion is also a valid criterion to consider speech as 'stuttering' in Dutch-speaking children (Boey, Wuyts, Van de Heyning, De Bodt, & Heylen, 2007). Stuttering severity was determined by administering the Stuttering Severity Instrument - 3 (Riley, 1994) to all children scoring above this 3% criterion. This instrument consists of 3 components, i.e. frequency, duration, and physical concomitants. For all of the participants, the nonreaders version was used to measure the stuttering frequency because of the delay in reading skills in children with DS (Næss, Melby-Lervåg, Hulme, & Halaas Lyster, 2012). Physical concomitants, such as distracting sounds and facial grimaces, were rated on a 6-point scale, ranging from 0 (none) to 5 (severe and painful looking).

Diagnostic criteria for cluttering

For this study, available cluttering checklists (e.g., Predictive Cluttering Inventory; Daly & Cantrell, 2006) could not be used. Therefore clinical characteristics frequently used in recent (working) definitions of cluttering were operationalized. St. Louis and Schulte's (2011) 'lowest common denominator' definition defines cluttering as

> A fluency disorder wherein segments of conversation in the speaker's native language typically are perceived as too fast overall, too irregular, or both. The segments of rapid and/or irregular speech rate must further be accompanied by one or more of the following: (a) excessive "normal" disfluencies; (b) excessive collapsing or deletion of syllables; and/or (c) abnormal pauses, syllable stress, or speech rhythm. (pp. 241-242)

Van Zaalen-op 't Hof (2009), focuses on similar main features, i.e.,

> a rapid and/or irregular articulatory rate, a higher than average dysfluency rate dissimilar to that seen in stuttering, reduced speech intelligibility due to bursts of fast speech and indistinct articulation, and telescoping, the merging of syllables with deletion of word parts within a word. (p.10).

In line with Coppens-Hofman et al. (2013), the following parameters important for the diagnosis of cluttering and differential diagnosis with cluttering-stuttering were evaluated: a) mean articulation rate (MAR) and variation of MAR (MAR-v), b) %OD and the ratio of OD and SLD, and c) telescoping. Additionally, both d) the frequency of silent pauses and abnormal pausing patterns, and d) speech intelligibility were evaluated. The latter two parameters were not part of Coppens-Hofman et al.'s diagnostic criteria for cluttering.

MAR and variation of MAR

MAR is based on three perceptually fluent measurement points of 5-10 syllables each (Coppens-Hofman et al., 2013) and is calculated by dividing the number of syllables in each

utterance by the duration (in seconds) of the utterance. These perceptually fluent utterances were selected from the beginning, middle and end of the speech sample if sufficient fluent utterances were present. When no fluent utterances were present at the beginning, middle and end of the speech sample, the available utterances were taken regardless of place of occurrence. Given the impact of pauses on MAR, these utterances could contain pauses of maximum 250 ms (Sawyer, Chon & Ambrose, 2008; van Zaalen-op ‘t Hof et al., 2009).

The variation of MAR (MAR-v), the difference between the highest and the lowest measurement of articulation rate, was also calculated. MAR values exceeding 5.2 syllables per second (sps) and MAR-v values above 3.3 sps or below 1 sps are considered deviant (for norms see van Zaalen & Winkelman, 2014, p.167). Deviant MAR-v values indicate that a speaker is not able to adjust the articulation rate to the complexity of the task.

%OD, %SLD, and ratio OD/SLD

Cluttered speech often presents with more disfluencies than normal speech. Coppens-Hofman (2013) considered the presence of 10 or more OD per 100 syllables an indication of cluttering. However, while the 3% criterion of SLD is a decisive parameter whether speech is considered ‘stuttering’ or not, the 10% criterion of OD is not. That is, if less than 10% OD per 100 syllables occur, speech can still be considered ‘cluttering’, for example, when the speech is produced with a highly increased MAR-v.

Coppens-Hofman et al. (2013) calculated also the ratio of OD and SLD for which they slightly adapted the procedures of van Zaalen-op ‘t Hof et al. (2009); values exceeding

2.7 are considered indicative of cluttering, values below 1 indicative of stuttering[1]. However, van Zaalen, Wijnen, and De Jonckere (2011) stated that these figures can only be used as a guideline and not a sole differential characteristic since in an earlier study with children and adults with fluency problems (van Zaalen-op 't Hof et al.), only 75% of the individuals who cluttered and 85.7% of the individuals who stuttered met these criteria.

Telescoping

Several authors named telescoping one of the characteristics of cluttering (Daly & Cantrel, 2006; St. Louis, Myers, Bakker, & Raphael, 2007; van Zaalen-op 't Hof et al., 2009; Ward, 2006). Telescoping or 'condensing' of words is the merging of syllables, resulting in a reduction of phonemes or syllables. For example, the 4-syllable word 'television' is reduced to the 3-syllable word 'tevision'.

Silent pauses

The above definition by St. Louis and Schulte (2011) mentions 'excessive normal disfluencies' (which also include silent pauses) and abnormal pausing patterns. Also Myers (2011) refers to a disrupted flow or timing of a message in cluttering as a result of insertions of "pauses or fillers*"* (p.154). In line with Ambrose and Yairi (1999) and Coppens-Hofman et al. (2013), silent pauses were not included in our classification system for disfluencies (Table 2) but were measured separately in PRAAT as one of the parameters for cluttering.

[1] In van Zaalen-op 't Hof, Wijnen and De Jonckere (2009) marginally different values are given, i.e., above 2.99 for cluttering and below 0.9 for stuttering.

There is some disagreement among researchers concerning the cut-off point for defining a silent pause (Lickley, 2015; Oliveira, 2002) and thresholds between 50 ms (Martin, 1970) and 250 ms (Goldman-Eisler, 1972) or higher have been used in research. In a recent study Eklund (2004) reported a cut-off point between 70 and 90 ms for silent pauses, in line with Levin, Silverman, and Ford (1965). In this study we have used the cut-off point of 80 ms, measured in PRAAT.

The presence or absence of abnormal pausing patterns was evaluated based on repeated listening to the participant's speech samples.

Speech intelligibility

Speech intelligibility was measured using Strand and Skinder's (1999; see also Button, Peter, Stoel-Gammon, & Raskind, 2013) 7-point scale: 1 = no noticeable differences from normal, 2 = intelligible though some differences occasionally noticeable, 3 = intelligible although noticeably different, 4 = intelligible with careful listening although some words unintelligible, 5 = speech is difficult to understand with many words unintelligible, 6 = usually is unintelligible, 7 = unintelligible. A substantial reliability has been reported using this type of interval scaling procedures (Schiavetti, 1992).

Results

Disfluencies in children with DS

Table 3 gives an overview of the group data for each of the nine disfluency categories used in this study. Similar to Ambrose and Yairi (1999) mean percentages per hundred syllables and relative frequencies (or proportions) are presented. Also the percentage of

children with DS exhibiting the different disfluencies is included. It is remarkable that the mean percentage of OD and SLD are similar, with a slightly higher percentage of SLD than OD. This is also apparent based on the relative percentages. Within the group however there is considerable variability since the standard deviations for both total SLD and total OD are above 75%.

When we divided our sample into two groups based on age (3;00-7;11y and 8;00-12;06y), an interesting pattern emerged. In the youngest age group (n = 11) the mean percentage of OD was 2.79 (SD = 1.66) and SLD 1.58 (SD = 1.13). In the oldest age group (n = 15) the mean percentage of OD was 1.98 (SD = 1.79) and SLD 3.00 (SD = 2.77). In other words, the youngest age group had proportionally more ODs while the oldest age group had proportionally more SLDs pointing to a developmental trend.

INSERT TABLE 3 ABOUT HERE

The breakdown in disfluency categories, both the mean percentages as the proportions, illustrates that for the five types of SLD, blocks are most frequent, followed by single syllable word repetitions, part-word repetition, prolongations, and broken words. A notable finding is that blocks are the most frequently occurring type of SLD, presented in nearly 54% of the speech samples. Within the group of OD, interjections were identified considerably more frequently compared to the other OD, and occurred in 69% of the participants. Revisions are the least frequently identified OD-category.

A clear majority of the participants presented with interjections, multisyllable word repetitions and prolongations. Single syllable word repetitions, blocks, phrase repetitions and part-word repetitions were observed in about half of the participants. Revisions only occurred

in the speech of about a quarter of the participants, and broken words were only heard in a few speech samples.

Stuttering in children with DS

INSERT TABLE 4 ABOUT HERE

Notwithstanding SLD (also prolongations and blocks) occurring in the speech of about half or more of the participants, only 8 participants (6 boys, 2 girls), i.e. 31% of the participant group, had three or more SLD per 100 syllables and were therefore classified as being children who stutter (CWS; see Table 4).

Subsequent analyses of stuttering severity in these eight participants is presented in Table 5. Five children had a mild stuttering severity and 3 were moderate. More detailed analyses of the subscores revealed that the frequency of SLD in this group ranged from 3 to 11 per 100 syllables. The average duration of the three longest moments of stuttering was 1043 ms (*SD* = 301 ms). In two CWS no physical concomitants were noticeable but in the other six there were, with scores ranging between 1 and 7. Detected physical concomitants were primarily facial grimaces, such as lip pressing and jaw jerking, but also head movements, such as forward movements and poor eye contact, and distracting sounds, such as noisy breathing.

INSERT TABLE 5 ABOUT HERE

Cluttering in children with DS

The average OD/SLD ratio for our participant group was 1.37 (*SD* = 1.19; see Table 4). MAR could not be calculated for four speech samples because there were insufficient long and fluent utterances in the speech samples. Mean scores for MAR and MAR-v were respectively 4.06 sps (*SD* = 0.69) and 1.55 sps (*SD* = 0.78). Based on a combination of % OD, % SLD, ratio OD/SLD, MAR, and MAR-v, Coppens-Hofman et al. (2013), according to the procedures suggested by van Zaalen-op 't Hof et al. (2009), classified speech as cluttering, cluttering with normal MAR, cluttering-stuttering, stuttering, or no fluency disorder. Criteria for 'cluttering' were: % OD > 10, % SLD < 3, ratio OD/SLD > 2.7, MAR > 5.2 sps[2] and/or MAR-v > 3.3 sps; in case MAR and MAR-v were not exceeding these values the label 'cluttering with normal MAR' was given. 'Cluttering-stuttering' was diagnosed with the following parameters: % OD > 10, % SLD >3, in combination with either a ratio OD/SLD > 2.7 and normal MAR and MAR-v, or a ratio OD/SLD between 1 and 2.7. The diagnostic label of 'cluttering-stuttering' was finally also possible with % OD < 10, % SLD >3, ratio OD/SLD < 1, MAR > 5.2sps or MAR-v > 3.3 sps. None of our participants met any of these five clustered classification criteria for diagnosing cluttering, cluttering with normal MAR, or cluttering-stuttering.

Both telescoping and abnormal pausing patterns were only seen sporadically in our participant group. Telescoping occurred in five children but in four of them below a frequency of 1%. A clearly distinguishable abnormal pausing pattern was only present in one child. The average frequency of silent pauses was 9.60 per 100 syllables (*SD* = 4.46).

[2] Coppens-Hofman et al. (2013) used 5.4 instead of 5.2. Van Zaalen and Winkelman (2014) provide Dutch norms for different age ranges: 5.2 sps for children, 5.6 sps for adolescents, and 5.4 sps for adults.

Speech intelligibility was noticeably diminished in most of the participants with an average score of 3.84 (*SD* = 0.73) on the 7-point scale used; this points towards speech that is intelligible with careful listening but with some unintelligible words present. Several phonetic and phonological errors, such as cluster reduction, deletion of end consonants, stopping and gliding were found.

In conclusion, based on our results, 31% of our participants could be identified with stuttering, none with cluttering or cluttering-stuttering, although we need to add that some of them showed characteristics that are linked to cluttering.

Discussion

This study investigated speech samples of 26 children younger than 13 years of age with DS. Disfluency types were described and categorized in SLD and OD and the speech samples were classified into stuttering, cluttering, a combination of cluttering and stuttering, and no disfluency disorder. Stuttering was determined based on three or more SLD per 100 syllables (Ambrose & Yairi, 1999) and subsequently the stuttering severity (Riley, 1994) was determined. Cluttering was identified by using the categorization scheme of Coppens-Hofman et al. (2013), based on %OD, %SLD, and ratio OD/SLD, MAR, MAR-v, and telescoping; additionally also silent pauses and speech intelligibility were evaluated.

Disfluencies in children with DS

Comparing the results of the present study to results of studies with typically developing children is difficult, as the speech and language acquisition of children with DS is often different and delayed compared to children without DS (Chapman & Heshketh, 2001; Dodd & Thompson, 2001; Kent & Vorperian, 2013; Miller, 1992; Rondal, 1995) and should therefore be interpreted with caution. Also, scrutinizing the types of disfluencies in other population groups, such as children with autism spectrum disorders (Scott, Tetnowski, Flaitz & Yaruss, 2014) revealed that these present with different patterns of disfluencies than do normally developing children.

Although Ambrose and Yairi's (1999) normative disfluency data were based on children aged between 2 and 5 years of age (mean = 3;00 for CWS and 3;03 for controls) and our particpants' age range was between 3 and 13 years (mean = 8;07), the fact that most children with DS have a delayed cognitive development resulting in a lower mental age (Weis, 2014) allowed us to compare findings, albeit with the earlier mentioned reservations. Ambrose and Yairi (1999) found on average 1.33 SLD per 100 syllables and 4.33 OD per 100 syllables in a group of typically developing young children (TDC). A recent dissertation by Polfliet (2014), analyzing disfluencies in sixty 3- to 7-year-old Dutch-speaking TDC, found 1.34 SLD and 2.64 OD per 100 syllables. Our group had on average nearly twice the amount of SLD per 100 syllables compared to both Ambrose and Yairi's and Polfliet's TDC (2.41 vs. 1.33/1.34) but fewer OD (2.33 vs. 4.32/2.64). The relative frequencies of SLD and OD are almost similar in our group whereas in the study of Ambrose and Yairi the proportion of SLD was significantly higher in the stuttering group (SLD = 0.66; OD = 0.34) and considerably lower in the control group (SLD = 0.24; OD = 0.76). Furthermore, the relative frequency of SLD in our study approaches more that of the stuttering group than that of the control group in Ambrose and Yairi's study.

If we focus on the different disfluency categories, it is surprising that of all SLD, blocks are the category that most frequently occurs and that it occurs in over half of all speech samples. They, however, occur in low frequencies in nearly all speech samples, except for two in which 12 blocks were counted for each speech sample. Both findings are surprising since repetitions, not blocks, are known to be the most common type of SLD in normally developing CWS and children who do not stutter (e.g., Ambrose & Yairi, 1999; Guitar, 2006). Blocks are known to be rarely occurring in fluently speaking children (Ambrose & Yairi). A longitudinal study in which children with DS are followed over a longer period of time may give insight in the increased occurrence of blocks, e.g. if this is a temporary trend, or not. An unpublished study (dissertation) in this series of studies showed a similar increased occurrence of blocks in the speech of adults with DS (Withaegels, 2009), a finding not consistent with Coppens-Hofman et al. (2013). The high frequency of interjections in the speech samples of individuals with DS was also found in other studies (Ambrose & Yairi, 1999; Coppens-Hofman et al., 2013; Dejoy & Gregory, 1985; Wexler & Mysak, 1982).

Stuttering in children with DS

The prevalence of stuttering is generally estimated at 1% (Bloodstein, 1995). Yairi and Ambrose (2013) showed in a recent publication that although the prevalence under age 6 is considerably higher than in later periods in life, the average lifespan prevalence might be somewhat lower. The prevalence of stuttering in our sample was 31%. In general, it is accepted that the prevalence of stuttering in individuals with DS is much higher than in a normal population and figures range between 15 to 45% (Devenny & Silverman, 1990; Van

Riper, 1971). The prevalence, however, is usually estimated for adults with DS or a mixed age group with DS. The prevalence of a group of young children was never yet estimated.

With regard to the male-female ratio we noticed a similar trend as in the typically developing population, i.e. more boys stuttered compared to girls. In our participant group of 14 girls and 12 boys, we identified two girls and six boys with stuttering.

In line with earlier reports on core stuttering characteristics in people with DS (Bray, 2003; Cabanas, 1954; Willcox, 1988), we found all categories clustered under SLD, i.e. prolongations, blocks, monosyllabic and part-word repetitions to be present in the our participant group. Also in six of the eight children identified with stuttering, secondary stuttering behaviors were observed, ranging from facial grimaces to head movements and distracting sounds, similarly to earlier findings reported by Bray (2008), Devenny and Silverman (1990), Preus, (1972), Schlanger and Gottsleben (1957), and Stansfield, (1990).

Cluttering in children with DS

We focused on three main aspects of earlier described definitions to identify cluttering, i.e. a too fast and/or irregular speech rate with an abnormal pausing pattern, a higher then normal proportion of normal disfluencies, and a reduced speech intelligibility, impacted by the merging of syllables (telescoping). Based on the classification criteria, as used by Coppens-Hofman et al. (2013), we were not able to label any of our participants as cluttering or cluttering-stuttering whereas the same authors in their study of adults with an intellectual disability found cluttering or a combination with stuttering to be present in 75%

of their participants; however, it needs adding that only 39% of their participants were persons with DS. In our view, this could be attributed to several possible factors:

1. It might be possible that the development of cluttering symptoms is still in its initial stages and that the disorder has not yet entirely developed into full-blown cluttering. Diedrich (1984) reported that onset of cluttering can be established at about 7 years and also van Zaalen and Winkelman (2009) stated that in children below 8-to-10 years of age, the (differential) diagnosis of cluttering can be very challenging. Moreover, in a group of children with intellectual disabilities, which have been shown to acquire speech and language more slowly than children without DS (e.g., Chapman & Hesketh, 2001) this might be an even more decisive factor.

2. Linked to the previous point of delayed and/or disordered language development, one of the difficulties in pinpointing some of the symptoms related to cluttering in our samples is that many of the speech utterances were rather short. However, it is important to point out that our MLU-findings (Table 1) are in line with previous publications showing that children with DS on average start using two-word sentences around 3 (Oliver & Buckley, 1994) to 4 or 5 years of age, and three-word sentences around 7 to 8 years of age (Rondal & Buckley, 2003). Moreover, children and adolescents continue to produce shorter and less complex utterances than would be expected based on nonverbal mental age (Martin et al., 2009). *This is also illustrated by comparing our MLU-data to a recent study in Flanders (Pareyn et al., 2016): while the MLUs in our study (between 3;03 and 12;06 years of age) ranged between 1.86 and 6.87 (mean = 3.28; SD = 1.17), in typically developing Flemish children this ranges between an average of 4.17 (SD = 0.27) for 3-year-olds to 7.00 (SD = 1.25) for 7-year-olds. A similar increase in MLU (albeit somewhat lower) with increasing age was also apparent in the findings by Rice et al. (2010) in a US sample.*

3. Lastly, while the developed classification criteria are a pioneering step towards a better objectification of diagnosing cluttering, they may not be applicable to all ages and/or

different client populations. For example, one can wonder if the criterion of more than 10% OD should be used to label the speech of young children as cluttering. It is known that the speech of young children (despite whether they stutter or not) contains more disfluencies than that of older children (Byrd, Logan & Gillam, 2012; Wijnen, 1990, 1991). Van Zaalen-op ‘t Hof et al. (2009) suggest to use the 10% criterion as a criterion to identify cluttering in speech samples of (normal) individuals older than 6 years of age. Tumanova et al. (2014), however, reported more disfluencies (OD) in the speech of pre-school age CWS than in those who do not stutter. They suggested 7%OD and/or 8% total disfluencies as a criterion to identify stuttering in the speech of young children. More in-depth investigation is necessary to conclude whether 10% OD is also a valid criterion to identify cluttering in the speech of children younger than 6 years of age. This can be a reliable cut-off for children with a normally developing speech and language acquisition, but may not be correct to be used to classify the speech of population groups with a disorder that has an impact on the speech and language acquisition, and possibly cognitive development.

Telescoping and abnormal pausing patterns, other characteristics typical for cluttering, were hardly observed in our participant group, respectively in five and one particpant(s). The former is not surprising since the mean word length was 1.40 syllables (SD = 0.07), and the shorter the words, the lower the likelihood of telescoping. For the Dutch language, as far as we know, no normative data for the mean word length in syllables are available. For the English language there are some studies published. Flipsen (2006) showed a developmental trend of increasing length (syllables per word) with age in typically developing children, which was not found in children with delayed speech acquisition. Taken together with findings by e.g., Dyson (1998) and Yaruss (2000), these studies suggest a developmental trend with values rising from approx. 1.06 syllables per word for very young

children to 1.46 for older children and adults. However, it needs to be stressed that these figures are for English and comparing data between languages is difficult (e.g., Nettle, 1998). While some languages only have a limited number of one-syllable words (thus a higher mean word length), other, more compact languages, have many short words, lowering the mean word length.

Furthermore, there is ample evidence for a correlation between excessive speech rate and phonological processes like syllable deletion or telescoping (e.g., Haug-Hilton, Shüppert, & Gooskens, 2011; Ward, 2006) and also almost none of our participants showed excessive speech rates (MAR) or abnormal speech rate variations (MAR-v). In line with Guitar's (2006) statement that the tendency to exhibit telescoping is low in people who stutter, none of the five participants with telescoping were identified as CWS.

Pauses are known to be part of fluent speech (e.g., Kowal, O'Connell & Sabin, 1975), so it is not surprising to observe that silent pauses are (one of) the most frequently occurring types of disfluency. They are not commonly included in existing classification schemes (e.g., Ambrose & Yairi, 1999; Tumanova et al., 2014) nor have they been used in the more detailed analyses of cluttering populations (e.g., Coppens-Hofman et al., 2013; van Zaalen-op 't Hof, 2009) nonetheless they were included in definitions of cluttering (see above).

Although some of our participants demonstrated characteristics typically linked with cluttering, interestingly enough these characteristics did not cluster together within the same participants. In other words, while speech intelligibility was impacted in almost all participants, some participants showed abnormal MAR and/or MAR-v rates, but no abnormal disfluency percentages or telescoping, while other participants e.g., showed telescoping but no abnormal MARs. On the other hand, this might not be so strange for a disorder that has

been described in the past as ‘a syndrome’ (St. Louis, Raphael, Myers, & Bakker, 2003), i.e., a specific set of correlated observable signs and symptoms. The key question then is how many of these signs and symptoms need to be present in order to correctly diagnose the syndrome. This becomes somewhat problematic when specialists themselves differ in opinion as to which are core behaviors, crucial to diagnosis, and which are peripheral (Ward, 2006). In a similar line of reasoning, Ward uses the term “cluttering spectrum behavior” (p. 150) to describe persons exhibiting cluttering characteristics but for whom a cluttering diagnosis may be less evident. Finally, also Van Borsel (2011) highlights that the recent recognition of cluttering being a multifaceted disorder, by no means clarifies the picture and “might result in an unusually high incidence of cluttering in DS” (p. 96).

This could also explain why the use of cluttering checklists, such as the Predictive Cluttering Inventory (Daly & Cantrell, 2006), might more easily label certain patterns of characteristics as cluttering in this population. At least, this seems to be corroborated by Van Borsel and Vandermeulen‘s study (2008) in which 96% of their participant group with DS was labeled as cluttering or cluttering-stuttering based on this inventory. But the question we currently still ask ourselves is: is this truly cluttering or is this a specific disfluency pattern, that, by the mere fact that it occurs in a person with an intellectual disability and speech-language problems resulting in impaired intelligibility (noticeable in most of our participants), is perhaps (mis)diagnosed as cluttering.

Additional considerations

Earlier we have addressed the possible impact of low MLU on the difficulties in detecting cluttering symptoms. However, it is most likely that this also impacts the

prevalence of OD and SLD, especially since it was found that over 70% of SLDs in children who stutter occur in utterances exceeding their mean length of utterance (Zackheim & Conture, 2003). Consequently, also the time of onset and/or recovery of stuttering is likely to be impacted since language development, and more in particular language deficiency, was found to be a risk factor for the onset of stuttering (e.g., Anderson, Pellowski, & Conture, 2005; Hubbard-Seery, Watkins, Mangelsdorf, & Shigeto, 2007) as well as the chances on recovery (Yairi & Ambrose, 2005).

The participant sample size was relatively small (n = 26) for this age range (3;03 – 12;06), which presumably contributed to the variability in the data (i.e., large SDs for both total SLD and total OD). While current findings provide (preliminary) data on disfluencies in children with DS, a domain only scarcely researched, future studies would ideally match these children to a control group of fluent or disfluent children with a similar mental age.

It should be noted that our speech samples were recorded in one speaking situation only, which is a limitation of this study. It is possible that speech in different situations generate more cluttering-like symptoms. It is, however, also necessary to consider the difficulty of getting young children with DS (often with a delay in speech and language acquisition) to talk in different situations. Situations other than spontaneous speech, as suggested by van Zaalen-op ‘t Hof et al. (2009), are reading and retelling a story. These are not achievable for most of the young children of this population group. Even though a narrative task elicits more SLD in children (Byrd et al., 2012), a spontaneous speech sample is believed to be sufficient (Conture, 2001; Hutchins et al., 2009).

In this study, the criterion for silent pauses was the cut-off point of 80 ms, in line with Eklund (2004) and Levin et al. (1965). The authors acknowledge that besides such an absolute measure there are also other ways of defining silent pauses. Crystal (2011; Crystal & Davy, 2013) for example, distinguishes four degrees of silent pauses: brief (the shortest category), unit (i.e. equivalent in length to one beat or cycle of the individual's normal rhythm of speech), double (twice as long as the unit pause), and treble (approximately three times as long as the unit). This subcategorization is not based on absolute numbers (fractions of a second) but on the person's own rhythm of speech. Eventually also such a classification system could be employed to analyze the specificity of silent pauses in this population.

Conclusions

Approximately 30% of the children with DS between 3 and 13 years of age were found to stutter, which is much higher than the prevalence in normally developing children. This study showed that the speech of children with DS has a different distribution of types of disfluencies than has the speech of normally developing children. Although different cluttering-like characteristics were found in the speech of young children with DS, none of them could be identified as cluttering or cluttering-stuttering.

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Table 1: *Participant and speech sample characteristics.*

Participant	Age in months	Gender	Number of utterances	Number of words	Number of syllables	Average number of syllables per utterance	Mean length of utterance (in words)
1	40	F	49	103	143	2.92	2.10
2	47	M	50	97	129	2.58	1.94
3	57	M	52	190	243	4.67	3.65
4	59	F	32	92	135	4.22	2.87
5	68	M	50	94	121	2.42	1.88
6	71	F	50	121	176	3.52	2.42
7	74	F	50	150	207	4.12	3.00
8	91	F	50	157	211	4.22	3.14
9	92	F	50	191	249	4.98	3.82
10	94	M	52	122	179	3.44	2.35
11	94	M	34	124	179	5.26	3.65
12	96	F	30	206	282	9.40	6.87
13	97	M	43	149	199	4.63	3.46
14	103	M	50	106	169	3.38	2.12
15	112	M	46	168	252	5.48	3.65
16	126	F	50	179	230	4.60	3.58
17	126	M	39	172	237	6.08	4.41
18	127	F	50	138	196	3.92	2.76
19	132	F	50	171	230	4.60	3.42
20	133	F	50	302	429	8.58	6.04
21	136	M	50	176	261	5.22	3.52
22	139	F	48	162	229	4.78	3.37
23	148	F	34	145	212	6.24	4.26
24	149	F	50	93	126	2.52	1.86
25	150	M	33	99	147	4.45	3.00
26	151	M	50	102	139	2.78	2.04

Table 2: *Classification system used to categorize disfluencies.*

Category	Description and example
Stuttering-like disfluency (SLD)	
Part-word repetition	Repetition of a part of a word. E.g., “Ba-ba-balloon.”
Single syllable word repetition	Repetition of a single-syllable word. E.g., “I go to… to… to school.”
Prolongation	Stretching of a sound. E.g., “Ssssssome porridge please.”
Block	Not producing a sound due to tension. E.g., “Can I have an … [tension] apple?”
Broken word	Not finishing a word during the act of talking without tension. E.g., “Let’s play a ga- [silence, no tension].”
Other disfluency (OD)	
Interjection	Filling of pauses with words like [uhm] before, in between or after (a) word(s). E.g., “I like this end [uhm] that.”
Revision	Breaking off a sentence and replacing it by a new one. E.g., “Can I go – shall we go home?”
Phrase repetition	Repetition of at least two words. E.g., “I went… I went home.”
Multisyllable word repetition	Repetition of a word that consists of at least two syllables. E.g., “My teddy… teddy is lost.”

Table 3: *Relative frequency, mean percentage (per 100 syllables), and percentage of participants exhibiting the different disfluency categories*.

Category	Relative frequency	Mean % (SD)	Participant %
Stuttering-like disfluencies (SLD)			
Part-word repetition	10.51	0.50 (0.69)	46.15
Single syllable word repetition	13.62	0.60 (0.63)	57.69
Prolongation	7.78	0.39 (0.35)	61.54
Block	18.29	0.87 (1.65)	53.85
Broken word	1.56	0.08 (0.20)	15.38
TOTAL SLD	51.75	2.41 (2.31)	
Other disfluencies (OD)			
Interjection	26.07	1.23 (1.43)	69.23
Revision	2.72	0.12 (0.22)	26.92
Phrase repetition	9.73	0.43 (0.56)	50.00
Multisyllable word repetition	9.73	0.54 (0.53)	69.23
TOTAL OD	48.25	2.33 (1.75)	

Table 4: *Overview of stuttering and cluttering parameters for all participants*

Participant	Age in months	Gender	MAR	MAR-v	%SLD[d]	%OD[d]	Ratio OD/SLD	%Telescoping[d]	%Silent pauses[d]	Abnormal pausing	Speech intelligibility
1	40	F	/	/	0.70	1.40	2.00	-	19.58	-	4
2	47	M	/	/	1.55	0.78	0.50	-	14.73	-	3
3	57	M	3.40	1.81	3.70[c]	2.88	0.78	-	13.99	-	4
4	59	F	3.58	1.07	0.74	1.48	2.00	-	8.15	-	4
5	68	M	/	/	1.65	4.13	2.50	-	18.18	-	4
6	71	F	3.42	1.22	1.70	5.68	3.33	-	13.64	-	4
7	74	F	4.30	1.71	1.45	2.90	2.00	-	8.70	-	4
8	91	F	3.48	0.56	0.00	0.47	0.00	-	2.70	-	2
9	92	F	3.23	0.54	2.01	4.82	2.40	-	13.65	-	5
10	94	M	3.11	0.74	3.35[c]	3.35	1.00	-	13.41	-	3
11	94	M	2.95	0.75	0.56	2.79	5.00	0.56	7.82	-	5
12	96	F	3.43	1.11	0.71	0.71	1.00	-	6.74	-	4
13	97	M	4.35	1.95	5.03[c]	3.52	0.70	-	10.05	-	4
14	103	M	4.83	1.60	1.18	2.96	2.50	0.59	2.37	-	3
15	112	M	4.58	1.03	6.35[c]	1.19	0.19	-	7.94	-	4
16	126	F	4.40	1.96	1.30	3.04	2.33	-	4.78	-	4
17	126	M	3.50	0.23	3.38[c]	5.06	1.50	-	10.13	-	3
18	127	F	4.24	2.12	1.02	0.51	0.50	0.51	7.14	-	4
19	132	F	5.20[a]	3.63[b]	1.74	1.74	1.00	-	6.52	-	4
20	133	F	4.42	1.81	4.66[c]	1.17	0.25	-	4.89	+	3
21	136	M	4.05	1.82	1.15	1.15	1.00	0.77	10.73	-	4
22	139	F	4.34	1.56	3.06[c]	6.11	2.00	-	8.73	-	4
23	148	F	5.42[a]	2.28	0.94	0.47	0.50	-	7.55	-	3
24	149	F	4.42	2.52	1.59	0.79	0.50	-	5.56	-	5
25	150	M	/	/	10.88[c]	1.36	0.13	-	14.29	-	5

26	151	M	4.70	2.06	2.16	0.00	0.00	2.16	7.91	-	4

/ : MAR and MAR-v could not be calculated

[a]: MAR> 5.2 SPS is indicative of cluttering (Van Zaalen & Winkelman, 2014)

[b]: MAR-v> 3.3 SPS is indicative of cluttering (Van Zaalen & Winkelman, 2014)

[c]: SLD>3% is criterion for stuttering (Ambrose & Yairi, 1999)

[d]: % is based on number per 100 syllables

-/+: characteristic is absent/present

Table 5: *Overview of the stuttering severity rating (SSI-3; Riley, 1994) for the participants who stutter.*

Participant	Age in months	Gender	Frequency score	Duration score	Physical concomitants score	Total	Pc	Severity
3	57	M	10	2	0	12	12-23	Mild
10	94	M	8	6	0	14	12-23	Mild
13	97	M	10	6	7	23	41-60	Moderate
15	112	M	12	6	5	23	41-60	Moderate
17	126	M	8	6	1	15	12-23	Mild
20	133	F	10	6	4	20	24-40	Mild
22	139	F	8	4	2	14	12-23	Mild
25	150	M	14	8	4	26	61-77	Moderate