Cluttering identified

Differential diagnostics between cluttering, stuttering and speech impairment related to learning disability

Yvonne van Zaalen – op 't Hof

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Broddelen geïdentificeerd

Differentiaal diagnostiek tussen broddelen, stotteren en spreekstoornissen gerelateerd aan leermoeilijkheden

(met een samenvatting in het Nederlands)

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CHAPTER One

Introduction and thesis outline

Introduction

In spontaneous speech no one is perfectly fluent. Even the most eloquent speaker suffers from speech failures every now and then. Probably, most of us make these mistakes more often than we actually want to. Different kinds of speech failures exist. For instance, we can insert pauses, or add interjections (‘well’) or meaningless sounds (‘uh’) to gain time. It is also possible to restart a sentence when we notice that it does not properly express what we intend to communicate. Word repetitions and “stumbling over one’s words”, or, technically, difficulties in realizing a word form, are also quite common. In response to failures like this, some people may say: “*Oh, damn, I am stuttering again”*. But such hesitations and sentence or word structure errors are not stuttering. Only when a person very frequently produces very many of these hesitations and slips of the tongue, in all kinds of *different* speaking situations, this may be indicative of a disorder, not stuttering but, arguably, cluttering.

For a long time, cluttering was the orphan of speech- and language pathology. After the German Kussmaul (1877) and the Austrian Weiss (1964) drew attention to this remarkable phenomenon, it was – in Europe in particular – recognized as a specific disorder. Cluttering did not fit in any other nosological class defined until then and remained poorly understood until the end of the last century. In the last century a diversity of symptoms were associated to cluttering. In the United States of America cluttering was not recognized as a disorder separate from stuttering, till, in the ‘90’s a handful of publications clarified the difference between stuttering and cluttering (e.g. St. Louis, Raphael, Myers, and Bakker 2003, 2007).
Cluttering is now generally characterized by three main features: (1) a rapid and/or irregular articulatory rate (Daly, 1993; Damsté, 1984; Dinger, Smit & Winkelman, 2008; St. Louis, 1992; St. Louis, Myers, Cassidy, Michael, Penrod, Litton et al., 1996; St. Louis, Raphael, Myers & Bakker, 2003; Weiss, 1964); (2) a higher than average frequency of normal (non-stutter-like) disfluencies, (Myers & Bradley, 1992; St. Louis, 1992,1996; St. Louis et al., 2003) and (3) reduced intelligibility due to exaggerated coarticulation (deletion of syllables or sounds in multi-syllabic words) and imprecise articulation (Daly & Burnett, 1999; Damsté, 1984; Dinger et al., 2008; Gutzman, 1893; Mensink-Ypma, 1990; St. Louis et al., 2003; St. Louis, Raphael, Myers & Bakker, 2007; Van Zaalen & Winkelman, 2009; Voelker, 1935; Ward, 2006; Weiss, 1964). A remarkable characteristic of persons who clutter (PWC), already described by Kussmaul in 1887, is that their speech production problems diminish

when they pay attention to their speech production or when they slow down their speaking rate.

Aetiology

Weiss's (1964) often quoted definition considers cluttering to be the verbal manifestation of an underlying central language imbalance. As ‘language’ as a cognitive faculty involves many components and processes, one may ask what an ‘imbalance’ in this system would entail. According to Myers (1992), the imbalance is to be viewed as a problem in synchronizing various components of the language system during utterance production: “*The propositions, pragmatic intent, meaning, sequencing, phrasing, timing, articulation, and orchestration*….” of speech and language functions “…*require synergy and synchrony of function and form*.” (Myers, 1992, p.175). When one or more of the concomitant functions go awry (e.g., speaking at a rate that is faster than the individual can handle, producing an intent or meaning that cannot be readily coded), symptoms of cluttering surface. According to Myers (1992), cluttering may ultimately be looked upon as a disorder of timing both for the production of speech and language units. It has been suggested by many researchers that this disorder is – ultimately – based in a neurological deficit. Alm (2007) proposes that the problem in adjusting speaking rate in PWC is due to an inhibition problem in the basal ganglia system. Further research on underlying neurolinguistic processes like the role of the basal ganglia circuits in cluttering is needed to confirm this hypothesis.

Prevalence

Pure cluttering is supposed to occur in 5-16% of disfluent speakers (Bakker et al., 2005; St.Louis & Mc Caffrey, 2005) and 21-67% of PWS also show cluttering characteristics (Preus, 1992). In the adult population 1-2 % is considered to be a disfluent speaker, whereas 5% of the children (aged 2-9 years) are diagnosed as disfluent. Cluttering may be more prevalent than the literature suggests, with cluttering and stuttering-cluttering almost as prevalent as stuttering (St. Louis & McCaffrey, 2005). The co-morbidity of stuttering and cluttering is high. Weiss (1964) was even doubtful of the existence of pure stuttering and argued that all stuttering is based on cluttering. Published prevalence and incidence rates for cluttering were not based on the current working definition of cluttering (St. Louis et al., 2007) and should therefore be used with caution.

Treatment

Systematic studies on the efficacy of therapy for PWC are virtually nonexistent in the literature. Possibly as a side effect of the diagnostic problems, there is little consensus regarding appropriate intervention techniques. The focus of therapy for people who clutter is on strategies to improve rate control and intelligibility, language production and monitoring skills (Daly, 1996; Mensink, 1990; Myers, 1996; Van Zaalen & Winkelman, 2009; Weiss, 1964; Ward, 2006). Less often cognitive restructuring and social changes (for instance, change of profession) are mentioned as part of intervention programs in cluttering (Daly, 1996; Winkelman, 1990; Van Zaalen & Winkelman, 2009). Changing behaviour, especially speech behaviour is very difficult. Therefore, treatment sessions with a cluttering client should be held frequently and at short intervals in order to be effective (Winkelman, 1990). Issues concerning treatment will not be addressed in this study, the focus will be on (differential) diagnosis and underlying neurocognitive processes.

Diagnosing cluttering

“One of the problems in diagnosing and treating cluttering is that it often occurs in conjunction with other disorders, some of which are speech-language based and others that are not” (Ward, 2006, p. 359). Differential diagnostics between cluttering and stuttering is difficult because these disorders have similar characteristics and often occur in conjunction with each other. Differential diagnostics in cluttering and stuttering has been based predominantly on subjective clinical judgments. Clinical judgment in the assessment of cluttering should be based on different aspects of communication and cognition, such as speech rate, intelligibility or fluency (Myers, 1996; Sick, 2004; St. Louis et al., 2003, 2007; Van Zaalen, Myers, Bennett and Ward, 2007; Ward, 2006).

Objective norms for speech and language characteristics and results on questionnaires developed especially for cluttering are needed to complement and support subjective clinical assessments. In addition to improving diagnostic reliability, it is assumed that by formulating objective criteria more light can be shed on neurolinguistic processes underlying different forms of speech disfluency.

Cluttering and language

The widely accepted working definition of cluttering by St. Louis, Raphael, Myers and Bakker (2007) describes cluttering as a fluency disorder characterized by rate abnormalities, but does not refer to language impairments in PWC. However, hypotheses stressing a central role of (high level) linguistic processes in cluttering have a long history. Weiss (1964, p.1) assumed that cluttering was the manifestation in speaking of a 'Central Language Imbalance', a disorder affecting all channels of verbal communication as well as some other, non-verbal skills. Earlier still, Freund (1952) and Luchsinger (1963) identified linguistic components in cluttering when they characterized the disorder as a 'dysphasia-like' disability. Grewel (1970) observed that cluttering is often found in children with a delayed speech and language development. The linguistic attributes of cluttering were also noted by Van Riper (1982) when he included linguistic anomalies next to articulatory rate variations in his "track II stuttering" (stuttering with a strong cluttering component). Damsté (1984) had a similar approach, when he described 'dysphasia-like cluttering'. However, despite the fact that these authors and more recent studies (St. Louis, 1992; Daly, 1992; Ward, 2004) also pointed to the importance of language difficulties in cluttering, research on the language skills of PWC has thus far not yielded more than vague and broad descriptions, such as "problems in retelling a story," (Mensink, 1990) or "a limitation in language formulation" (St. Louis, 1992) and "disorganized language formulation" (Daly, 1996).

It is assumed here that more knowledge on the language component in cluttering can be gained by studying the formulation skills of PWC at different speaking rates and underlying lexical complexity conditions. It is hypothesized that PWC do not exhibit a language disorder, but do exhibit (transient) language formulation difficulties that are induced and/or exacerbated by an abnormally high or a highly variable speaking rate (Myers, 1992). A language disorder can be defined as a disorder that affects all kinds of linguistic information processing, particularly both receptive and expressive tasks. By contrast, language formulation disturbances affect production only, and are reflected by specific types of disfluency: hesitations, interjections and sentence and word revisions. A detailed qualitative and quantitative comparative analysis of language formulating difficulties in PWC and unaffected controls is assumed to assist in clarifying the nature of the deficit underlying cluttering.

Cluttering and language/speech impairment related to children with learning disability

The disorder of cluttering provides us with an example of how much speech/language disorders and learning disabilities can have in common (Gregory, 1995). For Preus (1996), cluttering has more in common with learning disabilities (LD) than with stuttering. Daly (1986) claimed that decreased expressive language skills are common characteristics of children who clutter (CWC) or have learning disabilities. Many researchers contend that the overlap between cluttering and learning disability exists mainly with regard to problems in expression, reading aloud and writing (Daly & St. Louis, 1986; Mensink-Ypma, 1990; St. Louis, 1992; St. Louis, Myers, Raphael & Bakker, 2007 in Curlee & Conture, 2007; Tiger, Irvine & Reis, 1980; Ward, 2006; Weiss, 1964, 1968). Nevertheless, clear descriptions of the commonalities and differences of disturbances in language production in CWC and LD-children are lacking in the scientific literature. Research on specific aspects of language abilities in cluttering and speech impairment related to learning disability has been limited to merely mentioning problems in language production, as reflected predominantly in a high occurrence of disfluencies.

Brain imaging

Differential diagnosis of cluttering and stuttering is also important when disfluent participants are included in research projects. In brain imaging studies participants are often included on the basis of a stuttering diagnosis based on the percentage stuttered syllables or a Stuttering Severity Instrument-III score (Riley, 1994), (Blomgren, Nagarajan, Lee, Li & Alvord, 2003; Giraud, Neumann, Bachoud-Levi, Wolff von Gudenberg, Euler, Lanfermann & Preibisch, 2008; Smits-Bandstra & de Nil, 2007). In some studies the presence of other speech language disorders is used as an exclusion criterion (Neumann, Euler, Wolff von Gudenberg, Giraud, Lanfermann, Gall & Preibisch, 2003). In none of the brain imaging studies cluttering components are described, neither as inclusion nor as exclusion criteria. According to Preus (1992) 21-67% of the disfluent population displays cluttering symptoms. When a cluttering component in stuttering participants of brain imaging projects is neglected, it is possible that in comparing brain activation of stuttering participants to controls inconclusive results will be found.

Personal characteristics

Initiated by Weiss (1964) also various cognitive weaknesses, as well as personality traits are related to cluttering (e.g. poor concentration and attention span, reading disorders, writing disorders, unawareness of symptoms, restlessness and hyperactivity, impatience, superficiality, casual acceptance of life, lack of consideration of the consequences of a given act or for other people, and a short temper that is easily placated). Weiss' findings were based on clinical observations and not studied thoroughly. At present it is uncertain if these characteristics indeed are related to cluttering. It is possible that the (speaking) rate abnormalities in cluttering underly most of the above mentioned personal traits. Although I am aware that these characteristics are ascribed to cluttering, issues concerning non speech related behavioural aspects will not be addressed in this thesis.

Underlying neurolinguistic processes

When cluttering and stuttering can be differentiated (by for instance differentiating symptoms or different responses to intervention) it is reasonable to assume that underlying, neurolinguistic processes/deficits are also different. It is a challenge to describe the underlying processes/deficits as precisely as possible and in such a way that the observations (different symptoms, different responses to intervention) can be explained adequately and new testable predictions can be derived. Description of underlying processes/deficits should fit in knowledge of and models for normal production of spoken language.

Thesis outline

This study has two following objectives: (1) clinical, diagnostic classification of the syndrome of cluttering and particularly the difference in symptomatology between cluttering and stuttering and between cluttering and speech problems related to learning disability; (2) to contribute to a (neurolinguistic) model of cluttering that provides a coherent explanation for the observed symptomatology; and elucidates the difference between cluttering and stuttering.

(1) For that purpose diagnostic instruments used in stuttering assessment are adapted to cluttering and new assessment instruments to identify cluttering are designed and validated in a large group of disfluent speakers in the age range 6;6 – 50 years. Since in current clinical practice cluttering is diagnosed and differentiated from stuttering on the basis of subjective interpretation of articulatory rate (variations), type of speech errors and type and frequency of normal disfluencies, I will take these symptoms as a starting point. Consequently, it is hypothesised that objective measurements of articulatory rate, articulatory rate variation; type and frequency of disfluencies and errors in word or sentence structure will differ between persons who clutter (PWC) and persons who stutter (PWS). In determining norms for speech and language characteristics, a deeper understanding of some of the variables underlying different neurolinguistic processes of fluency disorders will be acquired.

Due to a lack of differential diagnostic criteria between cluttering, stuttering and language/speech impairment related to a learning disability, cluttering is often detected later in life, or not at all. This has the undesired result that therapy results are very limited and communicative skills of affected persons remain poor. It is assumed that in cluttering rate abnormalities manifest in language formulation disturbances and that these disturbances diminish when speaking rate is reduced or linguistic demands (complexity) decrease(s). By contrast, I hypothesize that the language production errors in LD-children will not be affected by rate or linguistic task in the same way.

(2) Exact objective diagnostic criteria for cluttering will guide formulating the characteristics of the underlying deficits. A functional MRI study showing differences between PWC and PWS will assist in corroborating the differences. A theoretical analysis of language production in cluttering will be given using Levelt's model of language production (Levelt, 1989).

Consequently, this thesis is divided in two parts. Part I addresses differential diagnostic characteristics of cluttering, stuttering and speech impairment related to a learning disability. Part II addresses underlying neurolinguistic processes in these disorders.

The first chapter of part I (Chapter 2) describes an empirical study aiming to set objective norms for differential diagnostic assessment of cluttering and stuttering symptoms, based on the three main characteristics of cluttering proposed by St. Louis, Raphael, Myers & Bakker (2003): (1) fast and/or irregular articulatory rate together with (2) errors in syllable, word or sentence structure and/or (3) a high frequency of normal disfluencies (not being stuttering). Objective measures are compared to the subjective clinical judgment made by expert fluency therapists. As a result of this work an assessment protocol differentiating between cluttering and stuttering was developed for use in further research on cluttering. Chapter 3 describes results of the Predictive Cluttering Inventory (Daly and Cantrell, 2006) of persons with fluency disorders (participants were children who clutter, stutter or had speech impairment due to a learning disability) in relation to the subjective and objective measurements described in Chapter 2. A revised version of the Predictive Cluttering Inventory checklist is validated to detect cluttering symptoms.

PWC experience difficulties in making themselves understood in conversations, but many are able to produce correct syllable and word structures in restricted situations (Weiss, 1964; Damsté, 1984; Bezemer et al., 2006; Ward, 2006; St. Louis et al., 2007; Van Zaalen & Winkelman, 2009). To produce intelligible syllable or word structures, the speaker must exercise appropriate levels of speech motor control. Results of the study described in Chapter 2 made clear that in order to differentiate cluttering from stuttering a validated test on speech motor control in stuttering and cluttering at word level was needed. In Chapter 4 the validity of an assessment instrument specifically designed to assess speech motor control at the word level, was tested. Such an instrument may enable the speech language pathologist to differentially diagnose the speech characteristics between PWC and PWS.

PWC differentiate themselves from PWS on speech motor control at word level. The question arose whether PWC exhibit language production disturbances comparable to persons with speech impairment associated with a learning disability. Chapter 5 describes to what extent disturbances in the fluency of language production of children who clutter might be comparable to, or differ from, those observed in LD-children. A tentative connection is made with the underlying processes of language formulation. It is hypothesized that an increase in normal disfluencies and sentence revisions in children who clutter reflects a different neurolinguistic deficit than that in LD-children.

In Part II the underlying neurolinguistic processes in cluttering and other disorders of fluency are described and placed into a model of speech and language production. Chapter 6 describes an fMRI study in which the findings of studies described in Chapter 2-5 will be confronted with brain activation data in persons diagnosed with either pure stuttering or pure cluttering while producing strings of multisyllabic words. In this fMRI study the question will be addressed whether PWC and PWS display different neurocognitive processes when performing speech tasks that call upon increasing demands on speech motor and linguistic skills.

Finally, in Chapter 7 cluttering is discussed within the framework of Levelt’s (1989) language production model. Underlying neurolinguistic processes are described in relation to articulatory rate. In the final discussion an answer is provided to the question if cluttering is a language based fluency disorder. The thesis ends with a general summary.

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PART one

Differential diagnostic characteristics

Chapter Two

Differential diagnostic characteristics between cluttering and stuttering – part one:

Speech characteristics of persons who clutter, clutter-stutter or stutter.

A slightly adapted version of:

Y. van Zaalen – op ’t Hof, F. Wijnen and P.H. DeJonckere (2009c)

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Abstract

Speech-language pathologists generally agree that cluttering and stuttering represent two different fluency disorders. Differential diagnostics between cluttering and stuttering is difficult because these disorders have similar characteristics and often occur in conjunction with each other. This paper presents an analysis of the differential diagnostic characteristics of the two disorders, and a proposal for distinguishing between the two in clinical settings.

The main goal of this article is to set objective norms for differential diagnostic assessment of cluttering and stuttering symptoms, based on the three main characteristics of cluttering indicated/identified by St. Louis, Raphael, Myers & Bakker [St. Louis, K.O., Raphael, L.J., Myers, F., & Bakker, K. (2003). Cluttering updated, The ASHA leader, *ASHA, 4-5*, 20-22]: a fast and/or irregular articulatory rate together with errors in syllable, word or sentence structure and or a high frequency of normal disfluencies (not being stuttering). In this article objective measures are compared to the subjective clinical judgment made by fluency experts. In other words, which characteristics can be found in the speech profiles of persons who were diagnosed as people who clutter or stutter?

1. Introduction

Speech-language pathologists generally agree that cluttering and stuttering represent two different fluency disorders. Whereas research into stuttering has increased markedly in the past century, studies on cluttering remain scarce. “One of the problems in diagnosing and treating cluttering is that it often occurs in conjunction with other disorders, some of which are speech/language based and others that are not” (Ward, 2006, p. 359). Differentially diagnosing between cluttering and stuttering is difficult because these disorders have similar characteristics and often occur in conjunction with each other. For example, Van Borsel and Tetnowski (2007) reviewed stuttering patterns in clients with mental retardation who showed evidence of disfluency patterns, concluding that not all would be considered stuttering. This paper presents an analysis of the differential diagnostic characteristics of the two disorders, and a proposal for distinguishing between the two in clinical settings.

Stuttering is a disorder characterized by a high frequency of involuntary interruptions of the forward flow of speech, regarded by the person who stutters (PWS) as “stutters”, which are often accompanied by a feeling of loss of control (Curlee & Conture, 2007; Guitar, 2006; Quesal, 2004; Shapiro, 1999; Van Borsel & Tetnowski, 2007; Ward, 2006). These interruptions usually take the form of (1) repetitions of sounds, syllables or one syllable words; (2) prolongations of sounds; (3) blocks of airflow or voicing in speech. The results of an exploratory study on speech motor practice and learning by Namasivayam and Van Lieshout (2008) indicated that PWS and persons who do not stutter may resemble each other on a number of performance variables (such as movement amplitude and duration), but they differ in terms of practice and learning on variables that relate to movement stability and strength of coordination patterns.

In the last decades of the previous century research on cluttering has addressed overt as well as covert symptomatology (Daly, 1996; Daly & Burnett, 1999; Myers & Bradley, 1992; St. Louis, 1992; Weiss, 1968). Weiss (1964) described cluttering as a disorder in the fluent flow of communication. According to experts, cluttering is characterized by three main features: (1) a rapid and/or irregular articulatory rate (Daly, 1993; Damsté, 1984; Dinger, Smit & Winkelman, 2008; St. Louis, 1992; St. Louis, Myers, Cassidy, Michael, Penrod, Litton et al., 1996; St. Louis, Raphael, Myers & Bakker, 2003; Weiss, 1964); (2) a higher than average frequency of disfluencies, dissimilar to those seen in stuttering [see section on stuttering characteristics above], (Myers & Bradley, 1992; St. Louis, 1992,1996; St. Louis et al., 2003) and (3) reduced intelligibility due to exaggerated coarticulation (deletion of syllables or sounds in multi-syllabic words) and indistinct articulation (Daly & Burnett, 1999; Damsté, 1984; Dinger et al., 2008; Gutzman, 1893; Mensink-Ypma, 1990; St. Louis et al., 2003; St. Louis, Myers, Bakker & Raphael, 2007; Van Zaalen & Winkelman, 2009; Voelker, 1935; Ward, 2006; Weiss, 1964).

1.1 Rapid and/or irregular articulatory rate

According to the St. Louis et al., (2003) working definition of cluttering a high and/or irregular articulatory rate is a main characteristic in differential diagnostics between cluttering and stuttering, however, agreement on what defines abnormally fast and abnormally irregular articulatory rate is needed. It is hypothesized that there are persons who clutter who maintain a high articulatory rate in a more demanding speaking situation, and their speech-language system can not handle such fast speed. Due to speech motor or language planning problems in a high articulatory rate, intelligibility problems or disfluencies occur (Daly, 1992).

1.2 Intelligibility and articulatory accuracy

Many researchers and clinicians (Bezemer, Bouwen, & Winkelman, 2006; Ward, 2006) report that people who clutter experience intelligibility problems due to exaggerated coarticulation (deletion of sounds or syllables in multi-syllabic words) and indistinct articulation (substitution of sounds and/or syllables). Several researchers discuss the fact that although persons who clutter experience intelligibility problems in running speech, they are able to produce correct syllable and word structures in controlled situations (Damsté, 1984; Van Zaalen & Winkelman, 2009; Ward, 2006; Weiss, 1964). The findings of Hennessey, Nang, and Beilby (2008) suggest that contrary to persons who clutter (PWC), PWS were not deficient in the time course of lexical activation and selection, phonological encoding, and phonetic encoding. In order to be able to produce correct syllable or word structures, speech motor control should be appropriate. Riley and Riley (1985) defined speech motor control as the ability to time laryngeal, articulatory and respiratory movements, that lead to fast and accurate syllable production. It is hypothesized that cluttering is a fluency disorder in which speech motor control at word level is disturbed in *high* speech rate, resulting in errors in word structure.

1.3 Frequency and type of disfluencies

Relying upon clinical experience, St. Louis, Hinzman, and Hull (1985) and St. Louis et al., 1996 differentiated the fluency disorders of disfluent people and concluded that PWC had a high frequency of normal disfluencies (e.g. revisions, interjections, phrase- and syllable repetitions) and a low frequency of disfluencies typical for stuttering. A higher than average frequency of disfluencies, dissimilar to those seen in stuttering is considered to be a characteristic of cluttering.

1.4 Subjective clinical judgment

Differential diagnostics in cluttering and stuttering has up till now mainly been based on the subjective clinical judgment of the speech-language therapist. Clinical judgment in the assessment of cluttering and stuttering should be based on different aspects of communication and cognition, for instance oral reading aloud, spontaneous speech, retelling a memorized story, and questionnaires (Sick, 2004; St. Louis et al., 2003, 2007; Ward, 2006). It would appear important to develop a more objective assessment method for the above mentioned aspects besides the subjective clinical judgment.

The main goal of this article is to set objective norms for differential diagnostic assessment of cluttering and stuttering symptoms, based on the three main characteristics of cluttering indicated/identified by St. Louis et al., (2003): a fast and/or irregular articulatory rate together with errors in syllable, word or sentence structure and or a high frequency of normal disfluencies (not being stuttering). In this article, objective measures are compared to the subjective clinical judgment made by fluency experts. In other words, which differentiating characteristics can be found in the speech profiles of persons who were diagnosed as people who clutter or stutter?

2. Method

2.1 Participants

All persons participating in this study had been referred to centres for stuttering therapy in the centre of the Netherlands (between January 2006 and January 2007) with self-reported fluency problems. Participants were 41 males (mean age 10.2; range 6–39 years) and 13 females (mean age 12.9; range 6-47 years). Controls for disfluent adolescents and adults were 17 males and 8 females (see Table 1.). A control group was included in order to obtain normative values for articulatory rate in retelling a memorized story and scores on speech motor control on word level in Screening Pittige Articulatie (SPA, van Zaalen Wijnen & Dejonckere, 2009a). For the tests used in the younger children no controls were needed, due to existing normative data. For the older participants, groups were matched for age and gender.

None of the participants (including controls) reported any neurological or hearing disorders and all were Dutch speaking mono- or bilinguals with an intermediate to high educational level. Subjects were tested in the first assessment session prior to therapy or in case of subjects that were already in the course of treatment (at the most 3 months) prior to any therapy session.

	Males			Females			Total
	N	Mean age	Range	*N*	Mean age	Range	*N*
Disfluent	41	10.2	6.0 – 39.4	13	12.9	6.3 – 47.2	54
Controls	17	24.3	12.6 – 47.3	8	25.2	12.4 – 52.1	25
Total	58			21			79

Table 1.: Participants divided in gender, mean age and age range

2.2 Diagnostic decision making

Participants were diagnosed based on subjective clinical judgment on audio recordings of three different speech tasks: spontaneous speech, reading aloud and retelling a story.

Diagnostic decisions were separately determined by two speech language pathologists specialized in fluency disorders (both cluttering and stuttering). Data was blinded and labelled in code. An independent researcher was in control of the coding system. Both speech language pathologists (SLPs) were aware of the age and gender of the participants. SLPs could choose between three diagnostic codes: cluttering (C), stuttering (S) and cluttering-stuttering (CS). A participant was appointed to a diagnostic group based on the diagnoses of both SLPs. When SLPs both diagnosed a person as cluttering, the participant was placed in the PWC group. When SLPs both diagnosed a person as stuttering, the participant was placed in the PWS group. When a participant was diagnosed as both cluttering and stuttering, he/she was placed in the PWCS group. When SLPs disagreed the participant was placed in the undecided group. After analysing the objective measurements of the PWS, PWCS and PWC group, participants in the undecided group were diagnosed based on the objective measurement in the three diagnostic groups.

2.3 Speech tasks

Data was gathered on: articulatory rate; articulatory accuracy and smooth-flow frequency and type of normal disfluencies. The test sequence for all participants was: (1) monologue; (2) reading aloud; (3) story retelling; and (4) speech motor coordination.

2.3.1. Task 1: Monologue

Participants were asked to recount an event in the recent past of their own choosing without intervention of the speech pathologist. Recordings lasted 3 - 5 min.

2.3.2. Task 2: Reading aloud task

This task and the next were adapted to the age / reading skills of the participants. Children read a standardised story, two levels below their reading level (as assessed in school). This was assumed to give the investigator a reasonable degree of certainty that not the reading skills but the speech skills were tested. The adults read a text above childhood reading level in order to examine complex sentences, multi-syllabic words and the appearance of more than one person in the story.

2.3.3. Task 3: Story retelling: the bus-story and the wallet-story

For the children under the age 12 we used "*the Bus story*" (Renfrew, 1997, Dutch version by Jansonius & Roelofs, 2006). This task is designed for use with children and has been used in several studies. Scoring norms are available. The researcher first tells the child the story while showing the child a colour book with 12 pictures of the story and accordingly the child is asked to retell the story. For the adolescents and the adults, we used the *Wallet story”*, which was adapted by Van Zaalen and Bochane (2007) from the ABCD test (Arizona Battery for Communication Disorders of Dementia).

In this study, an analysis of articulatory rate and disfluency measurements was undertaken. Due to lack of norms on this test, fluent age matched controls were also tested and means were compared.

2.3.4 Task 4a: Speech motor control on syllable level

Speech motor control can be measured with the Oral Motor Assessment Scale (OMAS, Riley & Riley, 1985). In a stable elicitation procedure ten repetitions of /puh/, /tuhkuh/ and /puhtuhkuh/ are obtained. These repetitions are judged on articulatory accuracy, smooth-flow (co-articulation, flow and sequencing) and rate.

2.3.5. Task 4b: Speech motor control on word level: the SPA test (Van Zaalen et al., 2009a)

Skills in oral motor coordination in multi-syllabic words were tested with the SPA test (Dutch: Screening Pittige Articulatie, Van Zaalen et al., 2009a). SPA, designed by the first author, is a specially designed speech task to provide information on speech motor control at word level in a fast speech rate. SPA can provide information on retaining correct word structure and intelligibility and was therefore included in our assessment procedure. In an elicitation procedure, three repetitions of ten complex multi-syllabic words at a fast speech rate were obtained. In order to allow a comparison with OMAS results, the SPA elicits three words containing (a) mostly bilabial onset consonants (similar to [pə]; e.g. Dutch: [ɔpərserəmɔ̀nimestər], or (b) mostly alveolar and velar onsets (as in [təkə], e.g. Dutch: [vərɑndərəndə ɭevənsɔmstɑndɪxhedən]; (c) a combination of bilabial, alveolar and velar consonants (as in [pətəkə], e.g. Dutch: [ɔnœytsprekələk vɛrveləndə vɛrhɑndəɭɪɳən]). These repetitions are judged on articulatory accuracy, smooth-flow (co-articulation, flow and sequencing) and rate.

2.4. Rate measurements

In order to obtain articulatory rate norms for fast rate, we needed an answer to the question: 'What is a fast articulatory rate?' To our knowledge, there is no clinical or scientific consensus on this issue. In disfluent speech, speech rate variation can be influenced by extra or extended pauses. Pindzola, Jenkins, and Lokken (1989) and Hall, Amir, and Yairi (1999) stated that articulatory rate measures are intended to reflect how quickly sound segments are produced in stretches of speech that have no pauses. We decided to base our judgment on Mean Articulatory Rate (MAR), which was defined as the mean of five rate measures in minimally 10 to maximally 20 consecutive syllables in perceptually fluent speech without pauses. We defined fast articulatory rate in spontaneous speech for disfluent speakers as a rate ≥ 1.0 SD above the MAR of disfluent speakers. It is well known that the fluent speech of PWS is slower compared to age and gender matched controls. The disfluent people are chosen as a reference group because it is known that differences in MAR between fluent and disfluent people exist.

Articulatory rate, in stuttering research, is usually calculated in syllables per second or phonemes per second by analysing only perceptually fluent utterances. Perceptually fluent utterances are defined as those utterances that exclude "within- or between-word disfluencies, hesitations, or pauses greater than 250ms" (Yaruss, Logan & Conture, 1994, p. 221).

In counting syllables one has to decide between the linguistic word form or the speech motor output. There are two reasons for choosing the linguistic word form in counting syllables of disfluent persons: (1) as cluttering is often considered to be a disorder of speech planning, it is important to know how much time the person planned to produce the word (Verhoeven, de Pauw & Kloots, 2004); (2) PWC sometimes produce unintelligible speech in which it is difficult to objectively determine how many and which syllables and phonemes have been realized. To avoid overly subjective assessment, the articulatory rate was calculated on the basis of the number of syllables that should have been realized [for the citations forms of words].

In the St. Louis et al., (2003) definition of cluttering deviant rate variability is considered to be a key symptom. For each subject, articulatory rate variability was computed as the standard deviation around the mean for the rate measurements. There are no normative values for MAR-variation (MAR-v). In our study, a deviant-MAR-v was defined as a variation in articulatory rate ≥ 1.0 SD above the mean articulatory rate variation: an indicator of cluttering.

2.5. *Ratio Disfluencies*

Evaluation of disfluencies, as a supplement to the diagnostic criteria based on articulatory rate and articulatory accuracy was done using Campbell and Hill's Systematic Disfluency Analysis procedure (Campbell & Hill, 1994). All disfluencies in samples of spontaneous speech and speech in story retelling were counted. The percentage of stutter-like disfluencies and normal disfluencies was calculated. The ratio disfluencies was obtained by dividing the percentage non-stutter disfluencies by the percentage stutter disfluencies, (for instance a participant with 20% non-stutter disfluencies and 2% stutter disfluencies had a ratio of 20/2=10) (Van Zaalen & Winkelman, 2009). It is expected that PWC will have a higher frequency of non-stutter disfluencies and therefore their ratio will be above one, while the person who stutters will experience more stutter disfluencies and their ratio will be below one. It is also expected that the mix group (PWCS) ratios will be either below or above one.

2.6. *Articulatory accuracy and coarticulation measurements*

In order to classify errors in sound, syllable or word structure, the Oral Motor Assessment Scale (OMAS; syllable level) and the SPA (Dutch: *Screening Pittige Articulatie;* word level) were applied. Scoring was done according to Riley's Oral Motor Assessment Screening protocol. Accuracy, smooth flow and rate were scored. Problems in sound accuracy (distortions and substitutions of voicing and devoicing) were scored. Accuracy scores on the OMAS or SPA more than 1.5 SD below the group mean were considered as an indication of severe problems in adjusting voicing to articulatory movement: an indicator of severe problems in realizing adequate voice-onset time. Problems in smooth flow were divided into three categories: coarticulation, flow and sequencing. Coarticulation is the gradual transfer from one speech movement to the other; errors in coarticulation include telescoping syllables and within sequence pausing. Flow is the gradual stressing and rhythm of the sequence; errors are for instance changes in stress pattern of the sequence. Sequencing errors are scored when a person makes errors in sound or syllable order. Smooth flow scores on the OMAS or SPA more than 1.5 SD below the group mean, are considered to be an indication of severe problems in oral motor coordination. Rate was determined by counting the mean time in seconds needed to produce three sequences of target syllables.

A reference test on oral motor coordination on syllable and word level is not available for ages over 12/13 years. In order to compare the results of the non-fluent speakers with fluent speakers, results of both adolescents and adults were compared to results of gender and age matched controls.

2.7. Diagnostic decision based on subjective and objective clinical judgment

The data of the participants with an undecided diagnosis were reanalysed, in order to determine the value of adding objective values to a subjective clinical judgment. Objective values obtained from the PWS, PWCS and PWC group, were used as an additional diagnostic component to diagnose the participants in the undecided group. For instance, when a participant was diagnosed as PWS by one SLP and PWCS by the other SLP and this participant scored according to the mean of the cluttering group (on ratio disfluencies or sequencing scores), the participant was diagnosed as a PWCS. In doing this, we could objectify the percentage of clients in which the cluttering component was missed by one SLP.

2.8. Data analysis

Recordings were made in a sound protected room. Digital audio- and video tape recordings were made of all speaking tasks using a Sony digital video camera, a Trust digital head microphone, and a GoldWave Digital Audio Editor v5.18.

Articulatory rate was determined using a speech analysis program, PRAAT (Boersma & Weenink, 2007), (see Figure 2). All of the fluent speech utterances produced by the individual subjects were recorded through a Trust head microphone into a high quality sound card using a HP Pavilion zv6000 laptop, sample frequency 22.050 Hz.

Digital audiotakes were edited, replayed and blindly analysed with PRAAT. The Mean Articulatory Rate (MAR) of five consecutive syllable strings was measured by counting the number of syllables per second in a string of at least 10 and maximum 20 consecutive syllables spoken, excluding pauses. Durational measures were made in ms by placing a cursor at the onset and another cursor at the offset. Onset was visually defined as the first peak (maximum amplitude in millivolts) that corresponded with a burst of spectral acoustic energy in the corresponding microphone signal or oscillogram. Offset was defined as the last consecutive peak in the waveform that was followed by a non-speech signal and also corresponded to the termination of spectral energy. Onset and offset of the utterance detected in the oscillogram and corresponding spectrographic display of each utterance were verified through playback of the auditory signal. Onsets and offsets of voiceless consonants that could not be clearly identified were excluded from the analyses, such as in the Hall, Amir and Yairi (1999) study. Next the duration of each utterance was calculated by subtracting the onset time from the offset time. Finally, the number of syllables for each utterance was divided by the duration of the utterance to provide a measure of articulation rate: syllables per second (SPS).

2.9. Reliability

A random sample of 8 participants was re-diagnosed to evaluate intra-judge reliability. A paired sample *t-test* between the two diagnoses was done ($p < .05$). In addition, a different random sample of 8 participants was re-analysed by the second experimenter, for evaluating inter-judge reliability on articulatory rate, articulatory accuracy and smooth flow measurements. In a paired sample *t-test* results obtained by the two experimenters were compared ($p < .05$).

	Diagnosis				
	PWC	**PWCS**	**PWS**	**Undecided**	Total
Subjective judgment	9 (17%)	10 (18%)	8 (15%)	27 (50%)	54 (100%)
Objective and subjective judgment	10 (18%)	23 (43%)	9 (17%)	12 (22%)	54 (100%)

Table 2.: Agreement on diagnostic decisions between speech-language pathologists based on subjective judgment or subjective judgment added with objective measurements.

3. Results

3.1. Diagnostic decision making

Pearson’s correlation between SLP diagnoses was low (r = .638). Of the 54 male and female disfluent speakers, only 27 (50%) were agreed upon by the SLPs. Of the 54 male and female disfluent speakers, 7 (13%) were diagnosed as PWC by one researcher and PWS by the other researcher; and 20 (37%) were diagnosed as PWC or PWS by one researcher and PWCS by the other researcher. Analyses presented here were carried out only on the 27 subjects that were agreed upon by the judges (PWC: N=9; PWCS: N=10; PWS: N=8) (see Table 2.).

3.2. *Articulatory rate*

Table 3 presents mean articulatory rate (MAR) in syllables per second (SPS) on three speech tasks (spontaneous speech, reading aloud and retelling) presented for each diagnostic group. Individual subject means for each speech task were calculated. In the case of two participants no individual mean articulatory rates could be calculated, as they were not able to produce at least 10 consecutive syllables.

No sex differences were found for MAR in monologue or retelling a memorized story, however, females were superior to males for reading aloud, [$F(1,22)=4.662$, $p= .047$]. Group differences were found for MAR in retelling a memorized story [$F(1,22)=8.489$, $p= .002$]. MAR of PWS was slower (SPS: *M*=3.7, *SD*=1.5) compared to PWC (SPS: *M*=4.9, *SD*=0.9) and controls (SPS: *M*=5.9, *SD*=0.5) (see Table 3.).

3.3 Fast articulatory rate

Fast articulatory rate (more than 1.0 *SD* above the MAR) was set at 5.5 SPS in monologue; 5.8 SPS in retelling a memorized story and 5.7 SPS in reading aloud. The majority of PWC (56%) met the description of fast articulatory rate in spontaneous speech, where PWS did not. No group differences were found for fast articulatory rate in reading aloud and retelling a memorized story.

	PWC (N=9)		**PWCS (N=30)**		**PWS (N=14)**		**Controls (N=25)**			
	M	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*	**F**	**Sig.**
MAR monologue	5.3	0.7	4.7	0.8	3.7	0.8				
MAR reading	4.5	1.7	4.6	0.7	4.0	1.0				
MAR retelling	4.9	0.9	4.8	1.1	3.7	1.5	5.9	0.5		
RD Monologue	6.4	3.9	3.2	5.2	0.4	0.5			8.7	.001**
RD Retelling	7.6	4.4	1.7	2.9	2.0	2.0			8.5	.001**
Accuracy SPA	2.1	1.7	1.6	1.3	0.2	0.8	0.1	0.2	11.4	.001**
Smooth flow SPA	8.7	2.0	6.8	2.2	1.4	3.2	0.8	0.9	38.4	.001**
Rate SPA	4.3	0.8	4.8	1.6	8.7	6.1	5.2	0.8		
MAR-v	2.5		2.2		2.4			0.1	.884	

Table 3.: Speech characteristics and between group analyses of variance, corrected by Tukey's b for unequal group size, on mean articulatory rate (MAR), ratio disfluencies (RD) and error scores on Screening Pittige Articulatie (SPA).

3.4. *Articulatory Rate variability*

No group differences were found for Mean Articulatory Rate –variation (MAR-v) (see Table 2). A deviant MAR-v was ≥ 3.99 SPS. A small number of persons who stutter (PWCS:23%; PWS:15%) fit the description of deviant MAR-v, while PWC did not.

3.4.1. *Ratio disfluencies*

Between group differences were found in ratio disfluencies for spontaneous speech [$F(1,21) = 34.787$, $p < .001$] and retelling a story [$F(1,21) = 16.874$, $p = .001$], but not in reading aloud [$F(1,17) = 3.171$, $p = .094$]. PWC produced 6.4 times more normal disfluencies compared to stutter disfluencies in spontaneous speech and 7.6 in retelling a memorized story. The Ratio Disfluencies for PWS was 0.4 for spontaneous speech and 1.2 for retelling a memorized story (see Table 3.).

A ratio of disfluencies above 2.9 is considered to be a cluttering symptom; whilst a ratio normal disfluencies below 0.9 is considered to be a stuttering symptom. In the PWC group 75% met the ratio disfluencies criteria for cluttering in both spontaneous speech and retelling a memorized story. In the PWS group 85.7% met the ratio disfluencies criteria for stuttering in spontaneous speech (see Fig. 1).

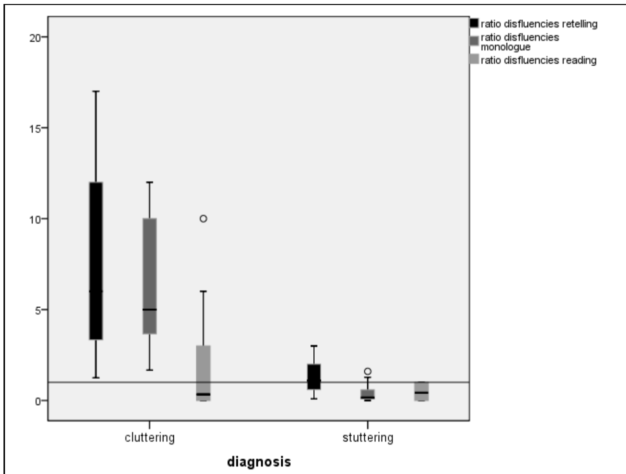

Fig. 1: Ratio disfluencies in the cluttering and stuttering group in retelling a story, spontaneous speech and reading aloud.

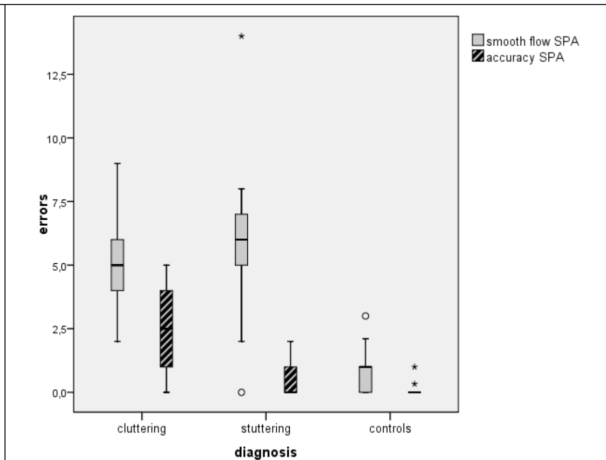

Fig. 2. Smooth flow and accuracy errors in Screening Pittige Articulatie (SPA)

3.5. Articulatory accuracy and smooth flow

PWC produced significantly more (*M*= 2.1) accuracy errors compared to controls (*M*= 0.19) and PWS (*M*= 0.21) in repeating multi syllabic word strings [$F(1,21)=11.386$, $p< .0001$]. Controls produced less smooth flow errors compared to PWS; PWS produced less smooth flow errors compared to PWC, [$F(1,54) =38.413$, $p< .0001$]. Smooth flow scores of PWCS were between PWS and PWC scores (see Fig. 2).

3.6. Adding objective measures to the subjective diagnostic decision making

When a ratio disfluencies > 2.87, reflecting a cluttering symptom, was added to the subjective clinical judgment 11 cases out of 54 could be added to the 27 cases SLPs agreed on diagnosis, with 29.6% of the disfluent cases still remain undecided. In adding accuracy problems > 2.1 (a cluttering component), to the subjective clinical judgment, the diagnosis of an extra 9 participants could be confirmed. In adding measures of both ratio disfluencies and accuracy error scores to the undecided diagnosis made by the subjective clinical judgment of the SLPs an agreement of 42 out of 54 (77,8%) diagnosis were agreed upon (see Table 2.).

3.7. Reliability

Intra-judge correlation coefficients on all metrics ranged between $.993 < r < .999$. Inter-judge reliability on articulatory rate, articulatory accuracy and smooth flow measurements ranged between $.675 < r < .868$.

4. Discussion

The main goal of this research was to compare the subjective clinical judgments made by experts in fluency disorders to results obtained by objective measurements. Findings indicate that a differential diagnosis based on a subjective clinical judgment of a speech-language pathologist specialized in fluency disorders appeared to correspond with the subjective clinical judgment of another SLP specialised in fluency disorders in only 50% of all disfluent cases. In 37.0% of all disfluent cases a client was diagnosed as PWC or PWS by one researcher and PWCS by the other researcher, in other words, one of the SLPs did not add the cluttering component to the diagnosis. Adding ratio of disfluencies and accuracy and speech flow error scores on word level to the subjective clinical judgment appeared to be of substantial diagnostic value, especially in locating a cluttering component. Of all cases, 15.2 % of diagnoses remained undecided after adding differentiating objective measures to the subjective diagnostic decision.

Overall, the rate data presented in our study are higher than those reported for (American) English-speakers. This finding corroborates data from earlier research where substantial rate differences among languages have been observed (Carlo, 2006; Grinfeld & Amir, 2006; Verhoeven et al., 2004). Articulation rate could be affected by linguistic as well as cultural aspects, thus the establishment of normative bases for mean articulatory rate for each language is essential. Fast articulatory rate for disfluent speakers, as defined as a rate ≥ 1.0 SD above the MAR of disfluent speakers, appeared to be of no differentiating value. One possible explanation of this result might reflect the decision to analyse only perceptually fluent or intelligible utterances.

In cluttering, a fast articulatory rate is mostly found in those utterances that are not fluent (in calculating SPS only fluent utterances were included) or in intelligible “spurts”. It would be important to consider these factors in future studies and find out a way to objectively measure such accelerated bursts of speech.

Sawyer, Chon, and Ambrose (2008) concluded, based on a single-speech sample in preschool children who stutter, that influences of rate, length, and complexity were not significantly correlated to stutter-like disfluencies. Contrary to that, a high amount of normal disfluencies in combination with a high level of syllable structure errors, can have a negative influence on the naturalness and intelligibility of speech (Levelt, 1989). Thus, cluttered speech that is perceived to be fast, may well be within normal limits when measured objectively.

Ratio disfluencies offer additional diagnostic criteria in retelling a memorized story. Based on the results of Boey, Wuyts, Van de Heyning, De Bodt, and Heylen (2007) we assume that results on the ratio of disfluencies can reasonably be used on both Dutch and English data. It is hypothesized that in retelling a memorized story a person who clutters does not adjust speech rate to the more complex language level resulting in a high level of normal disfluencies (word and phrase repetitions and interjections) and sentence structure errors (Van Zaalen & Winkelman, 2009; Van Zaalen, Wijnen & Dejonckere, 2009b).

Errors in speech motor control on word level and a ratio disfluencies above three, appear to be indicators for cluttering behaviour. Speech situations where a client is not focused on his/her speech or talks at a fast articulatory rate are sensitive to errors in speech motor control and disfluencies. In case of cluttering this will mainly be outside the clinic and in unstructured speech situations. Based on our results we advise SLPs to obtain data both outside and inside the clinic (both with the conscious knowledge of the client and not).

Although the present research provides ideas of setting normative data and procedures for differential diagnosis between cluttering and stuttering, the objective measurement values in this research are based on a small group of disfluent participants that both SLPs agreed upon. It is recommended that future studies on cluttering and stuttering include multiple factors or domains in the data collection process, especially with young children during the formative years of the disorder, when substantial overlap in the development of several speech/language domains occurs (Yairi, 2007), in order to better understand the intriguing disorder of cluttering.

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Chapter Three

Differential diagnostic characteristics between cluttering and stuttering – part two:

***Validation of the revised Predictive Cluttering Inventory*.**

A slightly adapted version of:

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Abstract

Speech-language pathologists generally agree that cluttering and stuttering represent two different fluency disorders. Differential diagnostics between cluttering and stuttering is difficult because these disorders have similar characteristics and often occur in conjunction with each other. The main goal of this second part of a two-part article is to discuss results on the Predictive Cluttering Inventory (Daly & Cantrell, 2006). Cluttering characteristics identified as diagnostically significant by 60 fluency experts. Proceedings of second world congress on fluency disorders are discussed in relationship to the subjective and objective measurements studied in the first half of the article and validate a revised version of the Predictive Cluttering Inventory.

1. Introduction

Van Zaalen, Wijnen, and Dejonckere (2009c) compared objective measures on articulatory rate, rate variation, type and frequency of disfluencies and scores on speech motor coordination tasks to the subjective clinical judgment made by fluency experts. This enabled the identification of speech characteristics that can be found in the speech profiles of persons who were diagnosed as people who clutter or stutter. In this article, results on the Predictive Cluttering Inventory (Daly & Cantrell, 2006) are discussed in relationship to the subjective and objective measurements studied in the first part of the article.

The Predictive Cluttering Inventory (PCI), (Daly, 1996; Daly and Cantrell, 2006) is a frequently used assessment tool. Daly and Cantrell (2006) called on 60 expert researchers and clinicians in cluttering worldwide to respond to a questionnaire containing a number of statements about the disorder. After analysing the data, the checklist contained 33 symptoms associated with cluttering, in four domains (pragmatics, speech motor, language and cognition & motor coordination and writing problems). Every symptom can be ranked with a score on a seven-point scale (0 = not present, 6 = always present) in order to predict possible cluttering. The PCI is produced without a norm for possible cluttering. The aim of the present study is to correlate PCI data with the characteristics of spontaneous speech production in disfluent and fluent speakers and validate the PCI as a cluttering detection instrument. For use in the Netherlands the PCI was translated into Dutch and back translated into English.

It is important to consider that the PCI is based on an email inventory developed by Daly and Cantrell (2006), where stuttering therapists around the world had to identify characteristics they assumed to be symptoms of cluttering. The 33-item PCI is the result of a factor analysis performed on all the characteristics named by the fluency experts. The PCI is produced without a discriminative norm, so it can be used as an inventory or checklist rather than a decisive instrument. Adding a discriminative norm could help SLPs in detecting cluttering more easily.

The PCI was developed to detect cluttering symptoms in running speech. A high sensitivity level is needed in order to accurately detect cluttering symptoms. A high level of sensitivity (> 75%) and a high level of specificity (> 75%) are needed in order to make the PCI a useful differentiating screening instrument. Children with learning disability (LD) were included, but while they have language disturbances similar to cluttering; they also produce a high frequency of normal (i.e., non-stutter-like) disfluencies, such as interjections, fillers, pauses, word- and phrase repetitions (Wigg & Semel, 1984). Fluent children with LD were hypothesized to score low on the PCI. The main purpose of this research was to investigate the validity of the PCI in a fluent and disfluent population. In other words is the PCI sensitive enough to detect cluttering in persons with cluttering symptoms and specific enough to reject people that do not clutter?

2. Method

2.1. Participants

In this study 137 Dutch speaking participants ranging in age between 10.6 and 12.11 years, were divided into five groups and examined by eight different SLPs. The groups were based on SLPs diagnosis as described in Van Zaalen et al., (2009c). Age range in all groups was restricted to exclude bias due to developmental issues. Group 1 consisted of cluttering children (*N*=17, *M*=11.5 years); Group 2 consisted of cluttering-stuttering children (*N*=25, *M*=11.6 years); Group 3 consisted of stuttering children (*N*=15, *M*=11.6 years); Group 4 consisted of children with learning difficulties (*N*=29, *M*=11.5 years); and Group 5 consisted of controls (*N*=51, *M*=11.2 years). None of the participants had known hearing or neurological problems. Disfluent children were recruited in two centres for stuttering therapy in the central and eastern parts of the Netherlands. Children that satisfied the inclusion criteria for age (from 10.6 to 12.11 years) at the time of the assessment in the therapy centres were included in the study. Children with learning disability and controls were recruited from a total of 21 (15 normal education; 6 special education) primary schools in Brabant (part of Holland). All schools have numbered class lists. In every participating school, six children were randomly selected on the basis of the student number on the class list. The parents received a consent form with explanation of the project prior to the assessment. Parents were asked for permission for participation of their children in the research project (informed consent). All parents gave permission for inclusion in the study.

2.1.1. Checklist

The Dutch translation of Predictive Cluttering Inventory, “Checklist Broddel Kenmerken” (CBK, Van Zaalen & Winkelman, 2009) was completed by a group of seven research assistants on the basis of observation of spontaneous speech, retelling a memorized story, reading aloud and parental information. All data was coded by an independent researcher who had access to the coding system. Researchers were SLPs with less than one year working experience. Completing the checklist was done by the researchers blinded (for SLP diagnosis). Groups were equally divided between researchers. In order to come closest to the use by SLPs around the world who download the checklist from the internet (the available source of the list), researchers were not informed on how to use the checklist other than by the notification made by Daly and Cantrell (2006) on top of the form.

2.1.2. Norm hypothesized by Daly (2008)

Daly (in press) hypothesized that a norm of 120 points in a 7-point scale (≥3 per item) would be sufficiently able to detect possible cluttering components in speech, based on clinical experience with cluttering clients. These hypothetical norms are based on subjective clinical observation by Daly and research is needed for their validation.

2.1.3 Reference test

While a ‘gold standard’ in cluttering assessment is lacking, we chose to use the subjective clinical judgment of two highly experienced SLPs specialized in fluency disorders, combined with objective measures (as described in part one of the article) as the reference test.

2.1.4. Analyses

Pearson correlations were used to determine relationships between subjective and objective clinical judgment to the checklist norm studied. Significant differentiating items

were analysed. Within group and between group differences were studied, using ANOVA. The sensitivity level, the proportion of actual positives correctly identified, and the specificity, the proportion of negatives which are correctly identified, were also determined.
A factor analysis was conducted to determine factors that together may explain the variance present in the basic variables. Results of the factor analysis were compared to results from a cluster analysis. The factor analysis grouping was based on the SLPs diagnosis (cluttering – non-cluttering), while in the cluster analysis, clustering of the items was conducted based on the individual item scores. In the hierarchical cluster analysis, between group linkages were displayed with squared Euclidean distance. It is hypothesized that items detected by both factor and cluster analysis present the best predicting cluttering items.

Items with significant differentiating value between diagnostic groups were found in the 33-item list by using an ANOVA with post hoc Tukey's-b correction for different group size and a significance level of $p < .05$. It was hypothesized that significant different items (for cluttering) with a mean value equal or above three (according to Daly's hypothesis) can serve as a predictor of possible cluttering. Supplementary to the total score of discriminating items, scores on factor one and cluster one items were studied. We hypothesized that a mean score above 3 (often – always) on all of these items provides a norm for cluttering.

Before using the checklist, items were not discussed between researchers to make sure that the use was similar to those therapists who download the file from the Internet. After completion of all checklists, all items were evaluated between researchers for the level of clarity. Item clarity analyses was done, by establishing inter judge correlation scores. Inter judge agreement levels were computed for two items on the checklist. Inter judge reliability level of >.70 was believed to be acceptable, meaning that both the item and the scoring system were interpreted similarly by different researchers.

After completion of all checklists, researchers could make comments on items they thought needed further explanation or clarification. Comments were taken into account in describing possible alterations of the checklist.

3. Results

3.1. Checklist

Based on the proposed norm score for cluttering by Daly (in press), only two participants from the cluttering or cluttering-stuttering group were detected by the checklist as having cluttering components (see Table 1.). That is, only 4.44% (2/45) of persons with cluttering symptoms were tested positive on the index test. This sensitivity level is considered to be extremely low. The specificity of the checklist is 94.7% (89/94), meaning, that 94.7% of a group of persons without cluttering indeed tested negative on the index test. The percentage of false positives, i.e., the percentage of positive tested people that did not clutter was high: 71.4%. The percentage of false negatives was high: 67.4%.

	Reference norm +	**Reference norm -**	**Total**		**Reference norm +**	**Reference norm -**	**Total**
Index test PCI +	2	5	7	**Index test PCI-r +**	22	9	31
Index test PCI -	43	89	132	**Index test PCI-r -**	10	86	96
Total	45	94	139	**Total**	32	95	127
Sensitivity	2/45= 4%			**Sensitivity**	22/32= 69%		
Specificity	89/94= 95%			**Specificity**	86/95= 91%		
False negatives	43/132= 33%			**False negatives**	10/96= 10%		

Table 1. Specificity and sensitivity of total scores on PCI and PCI-revised (PCI-r).

A factor analysis with a Varimax rotation on the cluttering item identified two factors with an eigenvalue above 1 explaining 85.8% of variance sums of squared loadings: Factor 1 included seven speech planning related items (loading: .61 - .86); factor 2 included six language structure items (loading: .61 - 83) (see Table 2.).

The cluster analysis with all 33 items on the list entered as variables identified four major clusters of coherent variables (see Table 3.). Cluster one contained those items involved in speech planning; cluster two contained items involved in language structure; cluster 3 contained items involving attentiveness and cluster 4 contained more common communicative skills.

Factor 1	Factor 2
Irregular speech rate; speaks in spurts or bursts	Language is disorganized; confused wording; word-finding problems
Rapid rate (tachylalia)	Disorganized language increases as topic becomes more complex
Telescopes or condenses words	Poor language formulation; poor story-telling; sequencing problems
Co-existence of excessive disfluencies and stuttering	Inappropriate topic introduction, maintenance, or termination
Initial loud voice trailing off to unintelligible murmur	Seems to verbalize before adequate thought formulation.
Little or no anxiety regarding speaking; unconcerned	
Oral diadochokinetic coordination below expected levels.	

Table 2. Items adjusted to factors 1 and 2 according to a factor analyses of the Predictive Cluttering Inventory (Daly & Burnett, 2006).

An analysis of variance between groups on total cluster scores corrected for uneven group size by Tukey's-b revealed significant differences (p= .01) between disfluent (cluttering and stuttering) and fluent groups (controls and LD) on cluster one: [$F(4,134)= 22.975$, $p< .0001$]; cluster two [$F(4,134)= 5.806$, $p< .0001$] and between controls and other diagnostic groups on cluster three [$F(4,134)= 12.961$, $p< .0001$]. There were no significant between group differences on cluster four.

A closer examination of these results in an analysis of variance on the clusters on mean cluster scores between disfluent speakers revealed no significant difference on the three clusters. While differences on total cluster score failed within disfluent speakers, both cluster two and cluster three showed significant differences between fluent speaking LD and controls (cluster two: [$F(1,78)= 12.146$, $p< .001$]; cluster three: [$F(1,78)= 25.230$, $p< .0001$]).

An analyses of variance between different diagnostic groups corrected for uneven group size by Tukey's-b procedure (p=.01) revealed six significant item scores on cluttering: irregular speech rate, speaks in spurts or bursts; rapid rate (tachylalia); initial loud voice trailing off to unintelligible murmur; little or no anxiety regarding speaking, unconcerned; co-existence of excessive disfluencies and stuttering**;** disorganized language increases as topic becomes more complex**.** The first five items mentioned above also appeared in factor 1 and cluster 1. The item "co-existence of excessive disfluencies" appeared in factor two and cluster two.

Cluster 1.: Speech planning	Cluster 2.: Disorganized language	Cluster 3.: Attentiveness
Lack of pauses between words and phrases; repetition of multi-syllabic words and phrases; irregular speech rate; speaks in spurts or bursts; telescopes or condenses words; initial loud voice trailing off to unintelligible murmur; oral diadochokinetic coordination below expected norm levels; co-existence of excessive disfluencies and stuttering; speech rate progressively increases (festinating).	Disorganized language increases as topic becomes more complex; poor language formulation; poor story-telling; sequencing problems many revisions; interjections; filler words; language is disorganized; confused wording; word-finding problems ; inappropriate topic introduction, maintenance, or termination; Improper linguistic structure; poor grammar; syntax errors	Does not recognize or respond to listener's visual or verbal feedback; does not repair or correct communication breakdowns; lack of awareness of own communication errors or problems; speech better under pressure; lack of effective self-monitoring skills; distractible; poor concentration; attention span problems

Table 3.: Distribution of items in cluster analysis.

3.2. Item clarity

Researchers were asked to comment on the comprehensibility of the checklist. Subjects and researchers should be able to comprehend the behaviors required to secure accurate and valid measures. One main concern (noted by all researchers) was uncertainty of how to interpret the scoring system. For instance, the word "often" can be interpreted as 'in almost every speaking situation' or 'in almost every sentence within a speaking situation'. Researchers interpreted this differently. This may have affected final scores.

Some items (item 3, item 6, item 21 and item 22) were considered difficult to score while a couple of conflicting symptoms were within one item description together. For instance, item 3 was difficult to score in case of conflicting scores on one part of the item description. Item 31 was difficult to score because oral diadochokinetic norms for adolescents and adults are not available and because it was not clear whether coordination on syllable, word level or during conversation was considered.

A checklist for possible cluttering should be highly sensitive on detecting cluttering. Therefore an adaptation of the PCI, based on the results of the present study was analysed. This involved selecting all the items that significantly differentiated cluttering from stuttering and controls (see Appendix A). A shortened version of the PCI was compiled on these eight differentiating items. A score of ≥ 3 on (24 points in total) all items of the shortened PCI resulted in an increase in sensitivity level to 69%. Specificity of the shortened PCI was very high at 91 % and the percentage false negatives was lowered to 10% (see Table 5.).

4. Discussion

The main purpose of this research was to investigate the validity of the PCI in a fluent and disfluent population. PCI/CBK in its original form is not sensitive enough to detect cluttering in persons with cluttering symptoms, but is specific enough to reject people that do not clutter.

The low sensitivity level of the PCI is equal to frequent type I errors, that is, a difference was observed when there was none. This may be explained by the fact that a lot of symptoms on the list are symptoms common to all disfluent speakers. For instance, every fluent speaker will produce more normal disfluencies compared to cluttering disfluencies; and weak time planning skills occur in people that do not clutter. Lack of adequate pausing is common in adolescent speech, but only considered a cluttering symptom when influencing speech intelligibility. Besides that, some items were expected to score high in the LD group. For instance, almost every child with learning disability will score high on attentional focusing problems and weak social skills. A few type II errors occurred, where there is a failure to observe a difference when in truth there is one. In case of cluttering, a type II error occurs if the test reports false when the person, in fact, clutters. The explanation of these errors is that some items are formulated in a way to differentiate cluttering from stuttering (items 8, 9, 19 and 20). It is expected that persons who only stutter will score low on this items but the difference between fluent speakers and PWC can be too small to be statistically significant. In solving these type I and type II errors, individual item scores were combined to cluster scores on a revised PCI (see Appendix A). The interpretation of item scores in the revised PCI heightened the sensitivity to a low but acceptable score of 69%. According to Pollit and Beck (2003), for group-level comparisons, coefficients in the vicinity of .70 are usually adequate. A sensitivity score of 70% is acceptable but due to the fact that some items and issues concerning the scoring system are not absolutely clear to all SLPs a short manual with clarification of items content and scoring system could further heighten the sensitivity of the PCI. The supporting symptoms are still in the revised PCI as these symptoms can be of great importance in therapy planning for an individual client (Bezemer et al., 2006; Daly & Cantrell, 2006; Ward, 2006).

In comparing the results of factor and cluster analysis, it was noticed that both analysing techniques came up with the same two main clusters: speech planning and language structuring. Another quality to consider in assessing this quantitative instrument is *speed*. Researchers were able to complete the PCI within 20-30 minutes. In a relatively short-time period the SLP is able to collect a substantial important data.

"One significant problem in trying succinctly to identify the characteristics of a clutter lies in the fact that there may be two basic strands to the disorder; a language component and a motor one" (Ward, 2006, p.141). The fact that it is common for cluttering to present more as a language problem than a motoric one, was supported by both factor and cluster analysis which proposed two major clusters of variables: a speech motor and a language component. "In case of linguistic cluttering speech output is more likely to show a lack of linguistic fluency, characterized by poorly constructed language rather than as an output which is motorically disrupted" (Ward, 2006, p.141), or as Daly described: "in cluttering accelerated speech is not always present, but an impairment of language formulation always is" (Daly, 1992, p.107). In cases of motoric cluttering speech output is more likely to show a lack of speech flow fluency characterized by excessive coarticulation, lack of speech rhythm, fast bursts of speech interspersed with short inappropriate pauses (Bezemer et al., 2006; Daly, 1996; Damsté, 1984; Dinger et al., 2008; St. Louis, 1992; St. Louis et al., 2003, 2007; Ward, 2006; Winkelman, 1990).

As described in the results section of item clarity, researchers reported that the scoring system was multi-interpretable. It is not clear whether scoring is done on one particular speech moment (for instance spontaneous speech) or concerns all speaking situations of a day (both focused and unattended speech) and what a particular score means ('always' = every day, every speaking situation or every word). It is known that speech and language disturbances of fluent speakers can differ between speaking situations and themes. In PWC the differences between speaking situations can be very large; especially the difference in speaking situations when attention to speech is given and those when a PWC is not alert to his/her speaking performance (Bezemer et al., 2006; Daly, 1992; Damsté, 1984; Dinger et al., 2008; Mensink, 1990; St. Louis et al., 2003; Ward, 2006; Weiss, 1964, 1968). This difference can partially be scored in item ten: *Speech better under pressure* *(improves short-term with concentration),* but sensitivity of the PCI would benefit from a more prominent place of this important symptom in cluttering.

Some item formulation more than likely needs to be reconsidered as many contain conflicting elements or researchers had problems in dealing with the negative statement in relation to the scoring system. However, using this revised scoring interpretation system produced improved sensitivity and specificity. While a number of type I errors still occurred in the revised PCI, it is not meant to be a diagnostic instrument on its own, though it has the potential to make contribution to the SLP working with disfluent persons.

In conclusion, the PCI in its current state does not serve as a valid diagnostic instrument for cluttering, but it serves as a valid screening instrument for possible cluttering symptoms. In its current state it does differentiate between fluent and disfluent speakers, but a differentiation between different fluency problems can not be based on total PCI scores only. Although the revised PCI subtotal score did not differentiate between cluttering and stuttering, the revised PCI could be of value in the prediction of cluttering components in speech. Further research on defining items is required in order to make this screening instrument a valid diagnostic tool that can be used by SLPs.

Appendix A

PREDICTIVE CLUTTERING INVENTORY (PCI)-revised
Original by Daly and Cantrell (2006); revised version by Van Zaalen et al., (2009)

INSTRUCTIONS to SLP: Please respond to each description section below. Circle the number you believe is the common most descriptive of this person's cluttering during the day. Count the scores of the itilized items in each section.

		5.Always	4.Almost Always	3.Frequently	2.Sometimes	1.Almost Never	0.Never
	Section 1: Speech motor						
1	*Lack of pauses between words and phrases*						
2	*Repetition of multi-syllablic words and phrases*						
3	*Irregular speech rate; speaks in spurts or bursts*						
4	*Telescopes or condenses words*						
5	*Initial loud voice trailing off to unintelligible murmur*						
6	*Oral diadochokinetic coordination below expected normed levels*						
7	*Rapid rate (tachylalia)*						
8	*Co-existence of excessive disfluencies and stuttering*						
9	*Speech rate progressively increases (festinating)*						
10	*Poor planning skills; misjudges effective use of time*						
11	Little or no excessive effort observed during disfluencies						
12	Poor planning skills; mis-judges effective use of time						
13	Articulation errors						
	Section 2: Language planning						
14	*Disorganized language increases as topic becomes more complex*						
15	*Poor language formulation; poor story-telling; sequencing problems*						
16	*Language is disorganized; confused wording; word-finding problems*						
17	*Many revisions; interjections; filler words*						
18	*Inappropriate topic introduction, maintenance, or termination*						
19	*Improper linguistic structure; poor grammar; syntax errors*						
20	Variable prosody; irregular melody or stress pattern						
	Section 3: Attentiveness						
21	*Does not recognize or respond to listener's visual or verbal feedback*						
22	*Does not repair or correct communication breakdowns*						
23	*Lack of awareness of own communication errors or problems*						
24	*Speech better under pressure (improves short-term with concentration)*						
25	Distractible; poor concentration; attention span problems						
26	Attention span problems;						
27	Seems to verbalize before adequate thought formulation						
28	Little or no anxiety regarding speaking; unconcerned						
	Section 4: Motor and planning (describe these symptoms compared to age level norms)						
29	Clumsy and uncoordinated; motor activities accelerated or impulsive						
30	Writing includes omission or transposition of letters, syllables, or words						
31	Poor motor control for writing (messy)						
32	Compulsive talker; verbose; tangential; word-finding problems						
33	Poor social communication skills; inappropriate turn-taking; interruptions						

Section one: > 24 points in itilized items => possible cluttering

Section two: itilized items provide supporting information on linguistic component in cluttering

Section three and four provide additional information on personal communicative skills

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Chapter Four

A test on speech motor control on word level productions: The SPA Test (Dutch: Screening Pittige Articulatie)

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Abstract

The primary objective of this article is to study whether an assessment instrument specifically designed to assess speech motor control on word level productions would be able to add differential diagnostic speech characteristics between people who clutter and people who stutter. It was hypothesized that cluttering is a fluency disorder in which speech motor control on word level is disturbed in high speech rate, resulting in errors in flow of speech and sequencing. An assessment instrument on speech motor coordination on word level was developed and validated. In an elicitation procedure, repetitions of complex multi-syllabic words at a fast speech rate were obtained from 47 dysfluent participants (mean age 24.3; SD 10.25, range 14.2–47.4 yrs) and 327 controls (mean age 25.56 yrs; SD 8.49; age range 14.3–50.1). Speech production was judged on articulatory accuracy, smooth-flow (coarticulation, flow and sequencing) and articulatory rate. Results from people who clutter (PWC) and people who stutter (PWS) were compared to normative data based on control group data. PWC produced significantly more flow and sequencing errors compared to PWS. Further research is needed in order to study speech motor control in spontaneous speech of people who clutter.

1. Introduction

Successful communication requires the active combination of a range of cognitive and linguistic skills. On the one hand, a speaker must coordinate a range of language-based faculties, including those required to competently formulate and structure sentences. Equally, speech planning and speech production processes are utilized to ensure that language-based elements are produced in an intelligible and coherent manner. Intelligibility itself is related to a number of factors, such as accurate sound production including speech rhythm, stress patterning, and articulatory rate.

It is a well known fact that persons with cluttered speech (PWC) experience problems in speech production resulting in unintelligible speech (Daly, 1992; St. Louis, Myers, Raphael and Ward, 2003; Weiss, 1964; Ward, 2006). It is hypothesized that cluttered speech occurs when speech rate is too fast for the speech system to handle, or when the person with cluttered speech does not give enough attention to the task of speech production.

Many researchers and clinicians report that PWC experience intelligibility problems due to exaggerated coarticulation (deletion of sounds or syllables in multi-syllabic words), indistinct articulation (substitution of sounds and/or syllables), and problems in accurate pausing (Bezemer, Bouwen & Winkelman, 2006; Daly & Cantrell, 2006; St. Louis, Myers, Bakker & Raphael, 2007; Ward, 2006). Several researchers discuss the fact that although PWC experience intelligibility problems in running speech, many are able to produce correct syllable and word structures in controlled situations (Bezemer et al., 2006; Damsté, 1984; St. Louis et al., 2007; Ward, 2006; Weiss, 1964). To produce intelligible syllable or word structures, the speaker must exercise appropriate levels of control over speech motor processes. Riley and Riley (1985) defined speech motor control as the ability to time laryngeal, articulatory, and respiratory movements that lead to fast and accurate syllable production. This ability is implicit in the widely accepted working definition of cluttering by St. Louis et al., (2007) which describes cluttering as:

A fluency disorder characterized by a rate that is perceived to be abnormally rapid, irregular or both for the speaker (although measured syllable rates may not exceed normal limits). These rate abnormalities further are manifest in one or more of the following symptoms: (a) an excessive number of disfluencies, the majority of which are not typical of people who stutter; (b) the frequent placement of pauses and use of prosodic patterns that do not conform to syntactic and semantic constraints; and (c) inappropriate (usually excessive) degrees of coarticulation among sounds, especially in multi-syllabic words. (p. 299).

In 1985, Riley and Riley published the *Oral Motor Assessment Scale* (OMAS). This instrument tests the ability of a speaker to produce intelligible syllable strings at a fast rate. Recent research (van Zaalen, Wijnen & Dejonckere, 2009c) revealed that adult and adolescent PWC performance on the OMAS cannot be differentiated from those of persons who stutter (PWS) or controls. PWC experienced no significant difficulties in oral motor coordination at the syllable level. Based on clinical observations in working with PWC, it is hypothesized that cluttering is a fluency disorder in which speech motor control at the *word level* is disturbed when speaking at a *fast* speech rate, resulting in errors in the flow and sequencing of speech.

The main purpose of this study is to test whether an assessment instrument specifically designed to assess speech motor control at the word level would be able to differentially diagnose the speech characteristics between PWC and PWS.

2. METHOD

2.1. Participants

All subjects in the PWC and PWS groups were referred to a centre for fluency therapy (between January 2006 and May 2008) in the centre of the Netherlands with self-reported fluency problems. Participants were 47 disfluent persons including 33 males (age: *M*=24.7, *SD*=9.8, range 14.4-49.3 years) and 14 females (age: *M*=24.3, SD=10.3, range 14.2-47.4 years) and 327 controls including 271 males (age: *M*=25.5, *SD*=7.9, range 14.1 – 54.3 yrs) and 56 females (age *M*=28.8, *SD*=8.5, range 14.3-50.1). Participants were divided in three diagnostic groups (PWC, PWS and controls). Diagnostic decision making was based on the objective results of the measurements on articulatory rate, ratio disfluencies, intelligibility, and the score on the Stuttering Severity Instrument (Riley, 1994). Diagnostic decision making procedures are described in detail in van Zaalen et al., (2009c). Controls were included in order to obtain normative values on speech motor control at the word level on the SPA test. None of the participants (including controls) reported any neurological or hearing disorders and all were Dutch speaking mono- or bilinguals with an intermediate to high educational level. Subjects were tested in the first assessment session prior to therapy or in case of subjects that were already in the course of treatment, (at the most 3 months) before the therapy session began. Adolescent and adult control subjects were selected at random from a database of volunteers originating in different parts of the country and who participated in the study. A total of 374 subjects participated in the study.

2.2. Speech motor control at the word level: The SPA Test

In order to examine the speech motor control on the word level, an assessment instrument was developed. The SPA (Dutch: *Screening Pittige Articulatie*), designed by the first author, is a specially created speech task that provides information on speech motor control and word structure productions at the word level when speaking at fast rates. In an elicitation procedure, three repetitions of ten multi-syllabic words at a fast speech rate were obtained. Stimuli were similar to those on the OMAS in that the SPA elicited three words containing (a) mostly bilabial onset consonants (similar to [pə]; e.g. Dutch: [ɔpərseremonimestər]), (b) mostly alveolar and velar onsets (as in [təkə], e.g. Dutch: [vərɑndərəndə ʅevənsɔmstɑndɪxhedən]); or (c) a combination of bilabial, alveolar and velar consonants (as in [pətəkə], e.g. Dutch: [ɔnœytsprekələk vɛrveləndə vɛrhɑndəʅɪɳən]). These repetitions were judged on articulatory accuracy, smooth-flow (coarticulation, flow and sequencing), and articulatory rate.

Articulatory accuracy and smooth flow measurements

In order to classify errors in word structure, the SPA (Dutch: *Screening Pittige Articulatie*) was developed (see Appendix 1). Scoring was devised to be consistent with Riley and Riley's (1985) Oral Motor Assessment Screening (OMAS) protocol. In this study, errors were defined within three different categories: (a) Accuracy, (b) Smooth Flow, and (c) Rate. Judgment of errors in sound or syllable production was based on a three point scale: Zero errors = 0 points, one to two errors = 1point, and three+ errors = 3 points. The more errors one produces, the higher the score.

2.2.1. Accuracy

Problems in sound accuracy (distortions or substitutions of voicing and devoicing) were scored. In the Dutch language, substitution of a target sound and the error may not have the same voicing category, as in English (i.e., [th] -> [s]). Accuracy scores for both PWS and PWC group that fell more than 1.5 SD above the mean score for the controls were considered to be an indication of problems in adjusting voicing to articulatory movement: an indicator of difficulty realizing adequate voice-onset time.

2.2.2. Smooth flow

Problems in smooth flow were subdivided into three categories: Coarticulation, flow, and sequencing. Coarticulation is the gradual transfer from one speech movement to the next. Errors in coarticulation were: telescoping syllables (i.e., Dutch: [ɔpsermonmestər] in stead of [ɔpərseremonimestər]), and within sequence pausing (i.e., Dutch: [ɔpərsere …. monimestər]). Flow is the gradual stressing and rhythm of the sequence. Errors, for example, may include changes in the stress pattern for the sequence (i.e., Dutch: [ɔpər<u>sere</u>monimestər] instead of [<u>ɔp</u>ərseremonimestər]). Sequencing errors were scored when a person makes sound order errors between or within syllables (i.e., Dutch: [ɔpərmoniseremestər] instead of [ɔpərseremonimestər]). Total smooth flow scores that are more than 1.5 SD above the mean score of the controlgroup were considered to be an indication of problems in speech motor control at the word level.

2.2.3. Rate

Rate was determined in syllables per second (SPS) by counting the mean time in seconds needed to produce a sequence of three target syllables. Normative comparison data was derived from the controls in this study. A reference test of speech motor control at the word level was not available. In order to get normative comparison data on accuracy, smooth flow, and rate, mean results of the non-fluent speakers were determined in z-scores.

3. RESULTS

3.1.1. Accuracy

A univariate analysis of variance between diagnostic groups corrected with Tukey's-b procedure for unequal group size revealed a significant group difference in Accuracy scores [$F(2,373)=66.675$, $p< .0001$]. Controls produced a mean of .23 (SD .53) accuracy errors. Controls produced significantly ($p < .0001$) fewer accuracy errors compared to PWC. PWC produced significantly [$F(1,46)=5.600$, $p= .022$] more accuracy errors compared to PWS (PWC: z-score *M*=1.65, *SD*=1.46; PWS: z-score *M*= .67, *SD*=1.23). (see table 1-3 and figure 1).

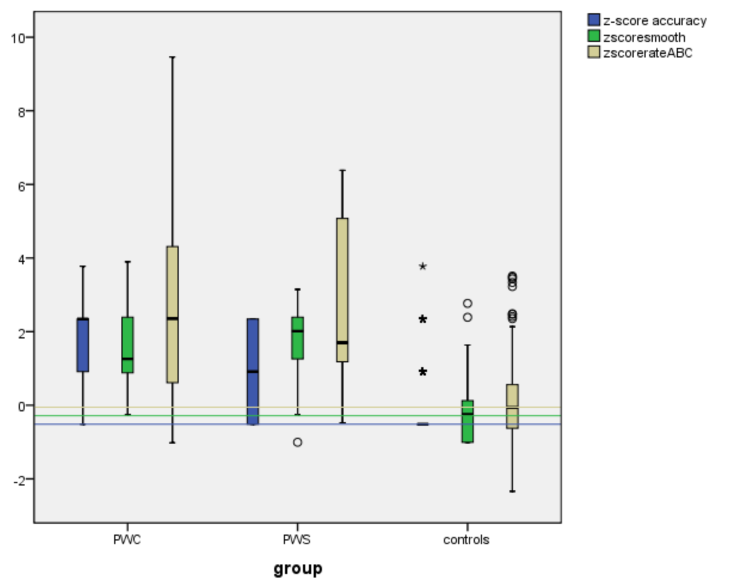

Figure 1.: Mean scores on accuracy, smooth flow and rate for PWC and PWS group and additional normative data and reference lines

3.1.2. Smooth flow

An analysis of variance between diagnostic groups on Smooth Flow errors revealed a significant group difference between controls and diagnostic groups [F(2,373) =116.460, p< .0001]. Controls produced a mean of 2.04, SD 1.91 smooth flow errors. Controls produced significantly (p < .0001) fewer smooth flow errors compared to PWC (z-score *M*=1.61, SD= 1.11) and PWS (z-score *M*=1.68, SD=1.20). Z-scores of PWS and PWC did not differ significantly [F(1,46)= .039, p= .844], (see Table 1-2). A closer examination of the smooth flow scores for the three different categories of coarticulation, flow, and sequencing was done.

3.1.3. Coarticulation

An analysis of variance between diagnostic groups on Coarticulation errors revealed a significant group difference [F(2,373) =88.959, p< .0001]. Both PWC and PWS produced significantly (p < .0001) more coarticulation errors compared to controls. (Coarticulation errors: Controls: *M*= .617, SD .97; PWC: z-score *M*= .63, SD= .68; PWS: z-score *M*=2.34, SD=2.22). PWS had significant higher Z-scores compared to PWC [F(1,46)=15.109, p < .0001] (see table 1-3 and figure 2).

	N	Minimum	Maximum	Mean	Std. Deviation	*F*	*Sign.*
accuracyABC	327	.00	3	.23	.53	66.675	0.00
smoothABC	327	.00	10	2.04	1.91	116.46	0.00
Coarticscore	327	.00	5	.62	.97	88.959	0.00
Flowscore	327	.00	3	.62	.72	58.428	0.00
Sequencescore	327	.00	5	.81	1.12	42.121	0.00
rateABC	327	3.1	8.5	5.2	.92	81.288	0.00
Passcore	327	3.8	16.5	7.5	2.35	194.267	0.00
Valid N (listwise)	327						

Table 1.: Normative values for Screening Pittige Articulatie

3.1.4. Flow

An analysis of variance between diagnostic groups on Flow scores revealed significant group differences [F(2,373)=58.428, p< .0001]. (Flow errors: Controls: *M*= .62, SD= .72; PWS: z-score *M*= .66, SD=1.33; PWC: z-score *M*=1.57, SD= .85). Both PWS and PWC produced significantly ($p < .0001$) more flow errors compared to Controls. PWC had significant higher Z-scores compared to PWS [$F(1,46)=8.079$, $p = .007$] (see Table 1-2 and Figure 2).

3.1.5. Sequencing

An analysis of variance between diagnostic groups on Sequencing errors revealed a significant group differences [$F(2,373)=42.121$, $p< .0001$]. Controls produced a mean of .807 (SD=1.12) sequencing errors (see Table 1.). Z-scores of PWS and PWC were significantly different, [$F(1,46)=4.782$, $p = .034$]. PWS (z-score *M*.41, SD 1.07) scored according to the controls. PWC (z-score *M*=1.42, SD=1.77) produced significantly more sequencing errors compared to controls. (see Table 1-2, Figure 2).

Group		N	Minimum	Maximum	Mean	Std. Deviation
PWC	Accuracy	29	-,52	3,78	1,6541	1,46190
	Smooth flow	29	-,25	3,90	1,6096	1,11368
	coarticulation	29	-,62	1,51	,6266	,67576
	Flow	29	-,90	2,62	1,5682	,85013
	Sequencing	29	-,71	4,16	1,4234	1,77168
	Rate ABC	29	-1,02	9,46	2,8257	3,18358
	SPA score	29	1,40	18,96	9,3678	4,68723
	Valid N (listwise)	29				
PWS	Accuracy	18	-,52	2,34	,6757	1,22667
	Smooth flow	18	-1,01	3,15	1,6775	1,19957
	coarticulation	18	-,62	5,77	2,3382	2,22438
	Flow	18	-,90	2,62	,6643	1,33520
	sequencing	18	-,71	2,07	,4100	1,07005
	Rate ABC	18	-,47	6,38	2,4800	2,09718
	SPA score	18	-1,97	16,97	8,2626	5,54493
	Valid N (listwise)	18				

Table 2.: Z-scores on subcategories (total error score on accuracy and smooth flow (coarticulation, flow and sequencing) and total rate in PWC and PWS groups).

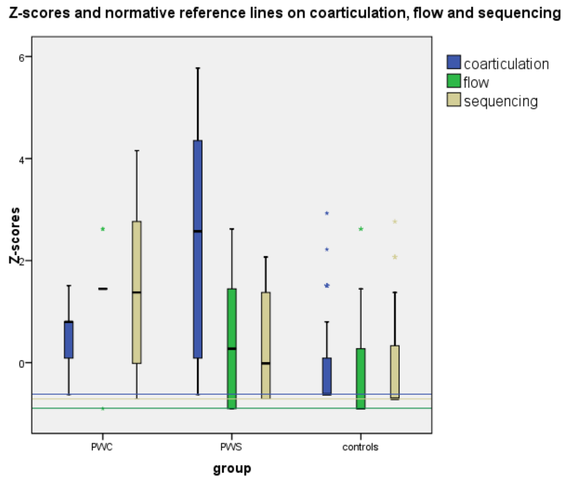

Figure 2. : Z-scores and normative reference lines on coarticulation, flow and sequencing

3.1.6. Rate

An analysis of variance between diagnostic groups on Rate scores revealed a significant group difference [F(2,373)=81.288, p< .0001]. Controls had a mean rate of 5.24, SD .92 seconds for the three words (see Table 1.). Both PWC and PWS had a significantly slower rate compared to controls(PWC: z-score *M* 2.83, SD 3.18 and PWS z-score *M* 2.48, SD 2.10). Z-scores of PWS and PWC were not significantly different, [F(1,46)=. 167, p = .685] (see Table 1-2).

3.1.7. Total SPA z-score

An analysis of variance on Total SPA z-scores between diagnostic groups showed a significant group difference. Controls had a significantly lower total score compared to PWC and PWS [F(2,371= 194.267, p < .0001]. Total SPA scores of PWC (z-score: *M*=9.4; SD=4.7) did not differ significantly from PWS (z-score *M*=8.3, SD=5.5), [F(1,46)= .536, p= .468]. (see Table 1-2 and Figure 3).

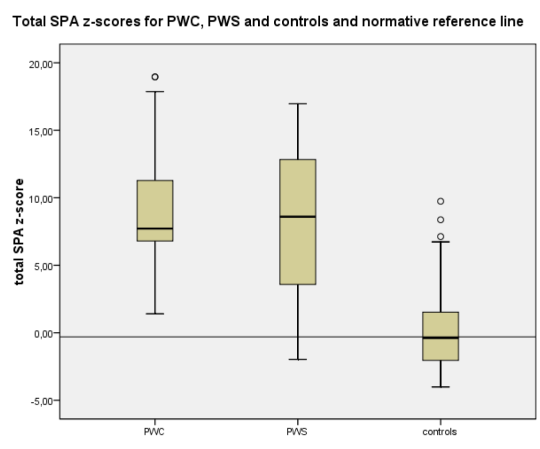

Fig. 3.: Total SPA z-scores for PWC, PWS and controls and normative reference line.

4. DISCUSSION

Many researchers have reported intelligibility problems in persons with cluttered speech (Bezemer, et al., 2006; Daly, 1996; Dinger, Smit & Winkelman, 2008; St. Louis et al., 2003; St. Louis, 2007; Ward, 2006). Until now, an assessment instrument for speech motor control at the word-level has not been validated for the disfluent population. The main purpose of this study was to test whether an assessment instrument specifically designed to evaluate speech motor control at the word level would be able to help differentially diagnose the speech characteristics of persons with cluttered speech and PWS. Results indicate that high scores on accuracy, flow and sequencing can differentiate PWC from PWS, but the total score on the Screening Pittige Articulatie (SPA) did not differentiate between PWC and PWS.

Accuracy scores

Accuracy scores on the SPA for the PWS and PWC groups with more than 1.5 SD above the mean of controls were considered to be indicative of problems in adjusting articulatory movement necessary to realize adequate voice-onset time. In this study, PWC had a mean z-score of 2.40 (SD=1.91) and met the criterion of severe problems in adjusting voicing to articulatory movement, while PWS did not. Problems in adjusting voicing can be seen when the timing demands for planning and execution processes can not meet (Howell, 2004). Target words used in the SPA, with complex phonetic and phonological properties that carry lexical stress, may cause more difficulty than those that do not have these properties (Howell & Dworzynski, 2005). This was evident for persons with cluttered speech.

Smooth Flow scores

Total Smooth Flow scores falling more than 1.5 SD above the group mean of the controls were considered to be an indication of severe problems in speech motor control at the word level. Total smooth flow scores appeared to be of no significant value in differentiating between PWC and PWS.

Coarticulation

In developing the assessment protocol, we replicated the major judgments categories used in the OMAS to the SPA. In the *Oral Motor Assessment Scale* (Riley & Riley, 1985) protocol, a major category named “coarticulation” contains both telescoping of syllables and extra pausing. Because PWS produced frequent extra pauses between and within words, their score on coarticulation was high. On the other hand, persons with cluttered speech produced frequent telescoping errors, while PWS rarely telescoped syllables. In combining pausing and telescoping data within the coarticulation category, potential difference might have been masked in the overall analysis. In future versions of the SPA, it is recommended that telescoping and pausing errors be split into two categories.

Flow and Sequencing scores

Scores on flow and sequencing errors differentiated the PWC from controls and PWS. Flow and sequencing abilities can be disturbed when speech planning has to be performed within small time limits. In the SPA, participants had to repeat test words at a fast speech rate. Goberman and Blomgren (2008) reported that stuttering speakers exhibited significantly more stuttering on variable rate tasks than on habitual rate tasks. Rieber, Breskin & Jaskin (1971) reported that PWS tend to have greater mean pause times and lower mean phonation times than persons with cluttered speech. In a fast rate, pause time is reduced, resulting in a higher frequency of flow and sequencing errors in the PWC group and a longer phonation time in the PWS group. PWS produced extra pauses that disturbed their flow of speech, but the syllable order was not disturbed.

PWC produced a high number of errors in a rate perceived to be fast, but not statistically different from the PWS. It is suggested that the accuracy and smooth flow problems in PWC negatively influenced their intelligibility. It is further suggested that articulatory rate in the PWC, although measured within normal limits, is perceived to be abnormally fast as a side-effect of other issues relating to problems in speech motor planning and speech motor execution (St. Louis et al., 2007; VanZaalen et al.,2009c). As Ward (2006) described, PWS seem to have problems producing what is already coded, while PWC experience problems in coding speech during conversation.

Total SPA score

The Total SPA Score was calculated by the summation of accuracy errors, smooth flow errors, and rate scores. The total SPA score for PWC and PWS was negatively influenced by speech rate. The total SPA score appeared to be of no value in the differential diagnosis within the disfluent population. This can be explained by the high scores for PWS in some categories and high scores for PWC in other categories. Both PWC and PWS experienced difficulties with speech motor skills.

Although this study has presented some new insights in the speech motor control in persons with cluttered speech or PWS, further research is needed in areas of spontaneous speech and other diagnostics groups that are related to speech motor planning or execution problems.

Speech motor control on word level

"Speech will be fluent if execution time for the segment currently being produced is sufficiently long for the plan for the following segment to be ready, after the current segment has been executed"(Howell & Dworzynski, 2005, p.352). Fluent speech needs separately planning and execution components (Levelt, 1989). Speakers can start an utterance (execution) before they have the complete plan (Kolk & Postma, 1997). When execution is getting ahead of planning, fluency problems arise. In testing speech motor on word level (multi-syllabic words) at a fast rate planning time was shortened. While PWC experienced accuracy, flow and sequencing errors as a result of that, it can be assumed that planning problems underlie on PWC production problems. While PWS experienced mainly coarticulation problems, it can be assumed that execution problems underlie on PWS production problems. Further research is needed to confirm this finding.

CONCLUSION

The main purpose of this study was to answer the question whether an assessment instrument especially designed to assess speech motor control at the word level would be able to differentially diagnose speech characteristics of PWC and PWS. Results show that smooth flow scores differentiate PWC from PWS. PWC produced significantly more flow and sequencing errors compared to the PWS. In addition, PWS produced significantly more errors on coarticulation compared to PWC and controls. Overall, the total score on the SPA test served to differentiate between fluent and disfluent participants. The SPA test on speech motor control on word level productions differentiated PWC from PWS.

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Appendix 1: Test protocol

Screening Pittige Articulatie (SPA)
van Zaalen, 2009

<u>Purpose:</u>
This assessment can produce insight in speech motor skills on word level within test circumstances. Of ten test words, only the three bold printed words are analysed on accuracy, smooth flow and articulatory rate.

<u>Instruction</u>*:*
The client can look at the word to produce during 5 seconds maximum. The word is covered and subsequently the client may repeat the words 3 times in consecutive syllable strings, in a fast but still intelligible way and without pauses.

Words (Dutch, bold words are test words):
periodieke uitkeringen = letterlijk en figuurlijk = veranderlijke wind uit westelijke richtingen = **onuitsprekelijk vervelende verhandelingen =** woordelijke aanhalingen = geldelijke tegemoetkoming = **opperceremoniemeester =** onverantwoordelijke elementen = **veranderende levensomstandigheden =** maatschappelijke verhoudingen

Name:

Mean number of errors in three attempts is determined and pointed out. Error score is pointed out in the row directly below.

	Accuracy						**Smooth Flow**								**Rate in SPS**
Word set	*Distorsion*			*Voicing*			*Coarticulation*			*Flow*		*Sequencing*			
Opperceremonie-meester	0	1-2	3+	0	1-2	3+	0	1-2	3+	yes	no	0	1-2	3+	Sec. A
Error score	0	1	3	0	1	3	0	1	3	0	1	0	1	3	
Veranderende levensomstandigheden	0	1-2	3+	0	1-2	3+	0	1-2	3+	yes	no	0	1-2	3+	Sec. B
Error score	0	1	3	0	1	3	0	1	3	0	1	0	1	3	
Onuitsprekelijk vervelende verhandelingen	0	1-2	3+	0	1-2	3+	0	1-2	3+	yes	no	0	1-2	3+	Sec. C
Error score	0	1	3	0	1	3	0	1	3	0	1	0	1	3	
Totaalscore															A+B+C = sec
Z-score															
Overallscore and interpretation															

Chapter Five

Language planning disturbances in children who clutter or have speech impairment related to learning disability

Y. van Zaalen – op ’t Hof, F. Wijnen and P.H. Dejonckere (2009b)

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Abstract
The primary objective of this paper is to determine to what extent disturbances in the fluency of language production of children who clutter (CWC) might be related to, or differ from difficulties in the same underlying processes of language formulation seen in children with learning disability (LD). It is hypothesized that an increase in normal disfluencies and sentence revisions in CWC reflect different neurolinguistic process to those of LD-children. To test this idea, 150 Dutch speaking children, age 10;6 - 12;11 years, were divided in three groups (cluttering, learning difficulties and controls), and a range of speech and language variables were analysed. Results indicate differences in the underlying processes of language disturbances between children with cluttered speech and those with learning disability. Specifically, language production of LD-children was disturbed at the conceptualiser and formulator stages of Levelt's (1993) language processing model, whilst language planning disturbances in CWC were considered to arise due to insufficient time to complete the editing phase of sentence structuring. These findings indicate that CWC can be differentiated from LD-children by both the number of main and secondary plot elements and by the percentage correct sentence structures.

1. Introduction

The disorder of cluttering provides us with an obvious example of how much speech language disorders and learning disability have in common (Gregory, 1995). For Preus (1996), cluttering has more in common with learning disabilities than with stuttering. Many researchers contend that the coherence of problems in cluttering and learning disabilities exists mainly with regard to problems in expression, reading and writing (Daly & St. Louis, 1986; Mensink-Ypma, 1990; St. Louis, 1992; St. Louis, Myers, Bakker & Raphael, 2007 in Curlee & Conture, 2007; Tiger, Irvine & Reis, 1980; Ward, 2006; Weiss, 1964). Daly (1996, p. 54) outlines the co-occuring features between cluttering and learning disability as follows: “*Children with the following symptoms: impulsive, disorderly, inattentive, under-achieving in school, specific reading problems and problems in language production, can easily belong to one of both categories*”. Decreased expressive language skills are common characteristics of CWC or LD-children (Daly & St. Louis, 1986; Tiger & Irvine, 1980). Nevertheless, clear descriptions of the specific disturbances in language production of CWC and LD-children are lacking in the scientific literature. Before outlining the research question, we begin by describing the characteristics of cluttering and learning disabilities. In addition, the similarities in language production deficits between these two populations will be clarified and discussed.

1.1. Cluttering

A widely accepted working definition of cluttering (St Louis et al., 2007, p. 299-300) describes the condition as follows:

> **Cluttering is a fluency disorder characterized by a speaking rate that is perceived to be abnormally rapid, irregular or both for the speaker (although measured syllable rates may not exceed normal limits). These rate abnormalities further are manifest in one or more of the following symptoms:**
>
> *a) an excessive number of disfluencies, the majority of which are not typical of people who stutter*
>
> *b) the frequent placement of pauses and use of prosodic patterns that do not conform to syntactic and semantic constraints; and*
>
> c) inappropriate (usually excessive) degrees of coarticulation among sounds, especially in multi-syllabic words.

This working definition does not refer to language impairments, which may be due to a lack of research into the linguistic components of cluttering. However, hypotheses stressing a central role of (high level) linguistic processes in cluttering have a long history. As far back as 1964, Weiss assumed that cluttering was the verbal manifestation of a central language imbalance, affecting all channels of verbal communication as well as other skills. Earlier still, Freund (1952) and Luchsinger (1963) identified linguistic components in cluttering when they characterized the disorder as “dysphasia-like”. The linguistic attributes of cluttering were also noted by Van Riper (1982) when he included linguistic anomalies next to articulatory rate as characteristics of his “track II stuttering” (stuttering with a strong cluttering component). But, despite the fact that St. Louis (1992), Daly (1992) and Ward (2004) also subscribed to this view on cluttering, research on the language skills of PWC has thus far not yielded more than vague and broad descriptions, such as “problems retelling a story,” (Mensink, 1990) or “a limitation in language formulation”(St. Louis, 1990). In view of this indeterminacy, a high frequency of normal disfluencies has come to be regarded the primary symptom of cluttering (Bezemer, Bouwen & Winkelman, 2006; Curlee & Conture, 2007; Damsté, 1987; Mensink, 1990; St. Louis, 1992; St. Louis, Raphael, Myers & Bakker, 2003; Ward, 2006; Weiss, 1964).

To summarize, research on specific aspects of language abilities in cluttering has been limited to merely mentioning problems in language production, as reflected predominantly in a high occurrence of disfluencies.

1.2. Learning disability

Children are diagnosed with learning disabilities if their ability to store, process or reproduce information leads to academic and or social problems due to a discontinuity in possibilities and achievements (Gettinger & Koscik, 2001; NCLD, National Centre of Learning Disabilities, 2002 Prior, 1996). LD-children have an IQ above 80 and deficits in at least one academic skill (reading, writing or mathematics). These difficulties are often associated with cognitive limitations, such as problems in short term memory and visual perceptual problems. Importantly, these discrepancies cannot result primarily from: (1) a visual, hearing, or motor impairment; (2) mental retardation; (3) emotional disturbance; or (4) environmental, cultural, or economic disadvantage (Council for Exceptional Children, 2004).

Most studies limit the description of language disturbances in LD-children (LD) to only mentioning descriptive data, while quantitative or normative data are lacking. In their study on language planning disturbances in children with LD Wigg and Semel (1984) observed a high frequency of normal (i.e., non-stutter-like) disfluencies, such as interjections, fillers, pauses, word- and phrase repetitions and syntax errors. To summarize, LD-children have deficits in at least one academic skill and often experience problems in language production, predominantly reflected in higher occurrence of normal disfluencies and syntactic problems.

1.3. Language planning disturbances

It can be hypothesized that similarities in speech disfluencies and language planning disturbances exist in CWC and LD-children. Up to now, the similarities and differences in language planning disturbances between CWC and LD-children have not been investigated. Understanding the nature of these impairments to language production may lead to a better understanding of the underlying processes in CWC or LD-children and to a more accurate differential diagnosis between these groups.

1.4. Levelt's language planning and production model and fluency

Language production models, especially Levelt's (1989) divide the process of preparing a spoken utterance into the following components: (1) conceptualising (creating the preverbal message; i.e. the speaker's intended message); (2) formulating (lexical retrieval, building of abstract linguistic representation – syntactic as well as phonological) and (3) articulating (transforming the linguistic representation to motor programming and production). Figure 1 reflects these components of Levelt's Model of speech production. The speech monitor detects and corrects speech errors. "*There are two ways of detecting errors: an internal loop, i.e. scanning the articulatory plan prior to its articulation; and an auditory loop, i.e. hearing one's own speech*" (Postma, 1991, p.114).

The disfluencies (interruptions and restarts) in this model are considered to be the result of difficulties at either the level of the conceptualiser or the formulator. The frequency of disfluencies reflects the degree to which phrases are withdrawn, reconsidered and mistakes were corrected (Levelt, 1999). Within Levelt's model, increased speech rate, either in bursts or at a more consistent speed, can result in excessive disfluencies because the speaker is unable to sufficiently organise the formulation process within the reduced time frame.

We hypothesize that the hesitations and disfluencies that LD-children experience are comparable to the hesitations and disfluencies of CWC. There are at least three different accounts of the cause of disfluencies for both LD children and CWC that we will describe below. Within this research project we want to investigate whether the cause of the disfluencies in CWC is different to that of LD-children.

Firstly, Postma and Kolk (1983, 1989), argue that hesitations reflect activity in the self-monitoring process. Speaking is interrupted as the result of the detection of an encoding error. This reactivity of the monitor is described by Postma and Kolk (1993) in their Covert Repair Hypothesis. Part word repetitions, prolongations and blocks are probably a result of covert repair in formulating processes.

Secondly, it is hypothesized that disfluencies can be caused by a (part of the) speech production plan that is not ready in time (Howell, 2004a; Kolk & Postma, 1997; Levelt, 1989). For example, it is possible that a word is not retrieved sufficiently quickly from the mental lexicon, or that constructing grammatical structures takes more time than normal. Disfluencies that result from this stalling (where a component of the production system fails to deliver output that can be processed by the next component) are typically seen in the emergence of pauses and initial syllable repetitions. It is possible that children produce these repetitions in order to gain time for organizing their language production (Howell & Dworzynski, 2005).The speaker can commence execution of the partly-prepared content word (Kolk & Postma, 1997) and use the remaining time to execute the remaining portion of the word or sentence. As the speakers use up the planning time, the plan may run out part way through the word (resulting in part-word disfluencies) or sentence (resulting in word- and phrase repetitions and interjections) (Howell & Dworzynski, 2005).

Thirdly, language production can also be perturbed by pragmatic problems. A concept can be clear at the moment the person starts to speak or express the message.

However, on the basis of the situation or the nonverbal information from the audience, a person can re-edit the whole plan (Clark & Brennan, 1991; Clark, 1997; Howell & Dworzynski, 2005; Smith & Clark, 1993). Revisions of sentences or content may result from this overt repair in story telling.

1.4.1. Conceptualizing and story telling

In the conceptualization phase of language production, story telling is dependent on the ability of the children to organise their thoughts into an overarching structure based on the primary plot elements and probably enriched with secondary plot elements (Renfrew, 1979).

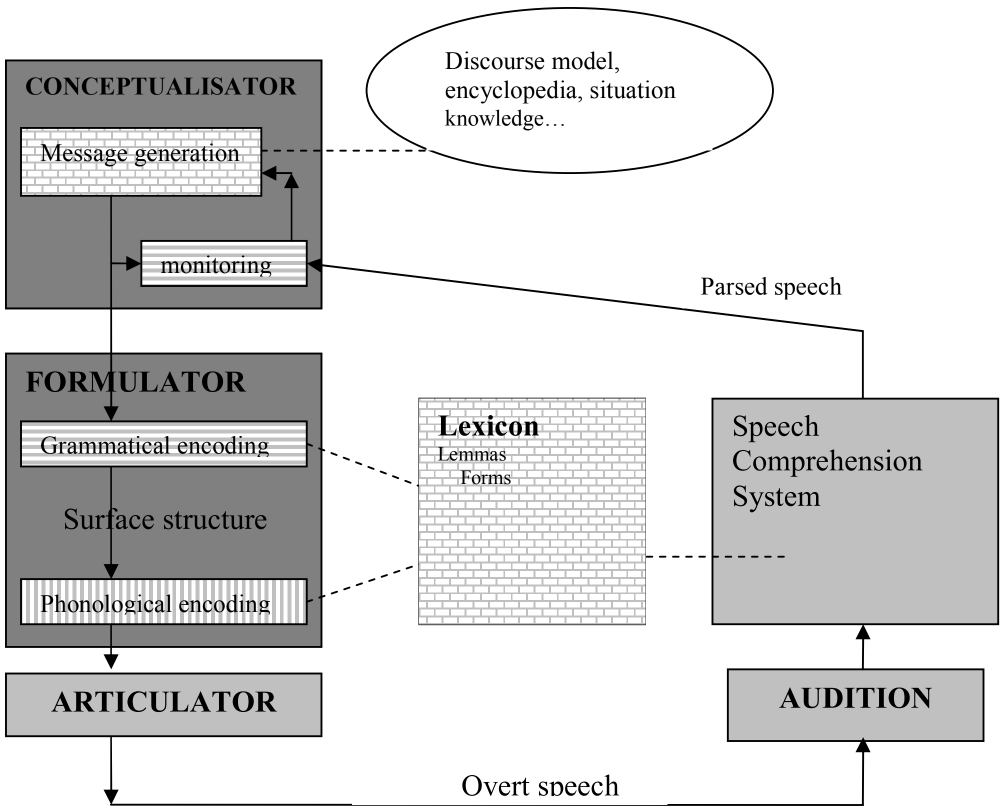

Figure 1.: Main language planning disturbances in LD-children (stones), CWC with problems in sentence structuring (horizontal lines) or CWC with problems in syllable structuring (vertical lines), after Levelt 1993.

It is assumed that when a child is not able to sufficiently reproduce the primary plot elements, problems exist either at the level of the conceptualiser (interjections and revisions) or with short term memory skills. Additionally, when a child produces a high number of hesitations during story telling, prior to complex words, the problems may lie at the level of the formulator, especially in lemma or word form retrieval.

1.4.2. Formulator and sentence structuring

Lexical selection drives grammatical encoding. Lemmas are retrieved when their semantic conditions are met in the message. In their turn, they activate syntactic procedures that correspond to the syntactic specifications resulting in correct sentence structures (Levelt, 1993). It is possible that a speaker makes an error in formulation that needs to be repaired. Making a self-repair in speech typically proceeds in three phases: detection, interruption and making a proper repair (Levelt, 1983). "*In self repair of a detected error or inappropriateness, interruption follows detection promptly, with the exception that correct words tend to be completed"* (Levelt, 1983). In editing of self repairs word and phrase repetitions are used as follows: "*After detection an error, or inappropriateness, the speaker will, in some way, 'transfer' structural properties of the original utterance to the correction* (Levelt, 1983, p.42). Levelt continued: "*By transferring and reusing structural properties of previous speech, (*during the editing phase)*, the speaker may at the same time gain in fluency*". Or as Levelt stated in 1994: "*During the editing phase of the formulation process, people repeat what was already formulated*" (Levelt, 1994).

In understanding the processes underlying hesitations and disfluencies in CWC or LD-children it is important to know to what extent these children are able to produce correct sentence structures. Based on the assumption that CWC or LD-children experience a high frequency of disfluencies, it is interesting to compare the sentence structures that are produced with, to those without, disfluencies. If the sentence structure was analysed both before and after the disfluencies were removed, important information could be garnered about the underlying processes of language production disturbances. And, in studying the different types of normal disfluencies, a better understanding of the conceptualiser and formulation skills of CWC or LD-children would be obtained. Improved possibilities for differential diagnostics in speech and language characteristics between CWC or LD-children could contribute to more efficient treatment planning of clients experiencing these disorders. The main purpose of this study is to answer the question whether disturbances in the fluency of language production of CWC are related to difficulties in the same underlying processes of language formulation as in LD-children.

1.4.3. Population

In selecting the research population it is important to consider that cluttering characteristics become recognizable when language acquisition and language production are in an advanced stage of development; that is, when children are capable of producing full fledged, grammatically correct sentences (Mensink-Ypma, 1990; Weis, 1964). Children in the last two or three years of secondary school in Holland (ages 10-13 yrs) satisfy this criterion. To reduce the influences of age on language production skills, it was important to limit the age range between the participants. Cluttering and learning disability can co-occur within a person (Curlee, 1996; Preus, 1996; St Louis, 1992; Weiss, 1965). The goal of this project is to study the similarities and differences in language production disturbances, therefore, disfluent LD-children were excluded from the study.

2. METHOD

2.1. Participants

In this study 103 Dutch speaking children ranging in age between 10.6 – 12.11 years were divided into three groups: Group 1: cluttering children (n=11, mean age 11.5 yrs); group 2: LD-children (n=37, mean age 11;6 yrs); and group 3: controls, n=55, (mean age 11;2 yrs).

Children with learning disability

LD-children were recruited from 21 primary schools in the province of Brabant (the Netherlands). Of 121 schools that were invited, only 21 agreed to take part in the study. Of the 21 schools, six provide special education for LD-children. In Holland, all schools have numbered class lists. In every participating school, six children were randomly selected on the basis of the class lists. When children in schools for special education did not meet the characteristics of learning disabilities as described below, another child in the same class was randomly selected.

All children that satisfied the criteria of learning disabled were included in the study. Learning disability in this study was diagnosed when (a) LD-children were classified as having learning disabilities by a multidisciplinary team according to Dutch special education regulations. (b) children had problems in at least one academic skill (i.e. reading, spelling or maths) associated with cognitive limitations and an IQ above 80. Due to cognitive limitations these children were not able to catch up in development with their peers and placement in a special education school was necessary. The IQ level was determined prior to placement in a school for children with special educational needs. (c) children had been enrolled in special education schools for more than three years, because of reading or spelling problems.

Only children who joined grade eight of the school for special educational needs were included in the study. Their reading level was comparable to the reading level of children in the fourth or fifth grade in normal education. LD-children who (according to either the teachers, the school speech-language pathologist (SLP) or the parents) stuttered were excluded from the study. In strictly applying the above mentioned criteria we tried to form as homogeneous a group as possible.

Children who clutter

Disfluent children were recruited in two centres of stuttering therapy in the central and eastern parts of the Netherlands. All children that satisfied the diagnostic criteria of cluttering were included in the study. Cluttering in this study was diagnosed when (a) the Stuttering Severity, determined with Stuttering Severity instrument-3 (Riley, 1994) was: no stuttering or extremely light stuttering; (b) the percentage stutterlike disfluencies was > 3.0; (c) the child spoke with a speaking rate that was either too fast or irregular, together with at least one other symptom of St. Louis's et al., (2007) working definition of cluttering.

Controls

Age matched controls were included in the study to provide information on the frequency and type of disfluencies in age matched, typically developing speakers without learning disability. In current scientific literature these data are missing for this group of children.
None of the participants had known hearing or neurological problems. All participants spoke Dutch as their mother tongue and had a normal level of intelligence. The first author provided a Dutch translation of the Predictive Cluttering Inventory (PCI, Daly & Cantrell, 2006) the Checklist Broddel Kenmerken (van Zaalen & Winkelman, 2009). This was sent by email and received prior to the assessment.

Informed consent

Parents received a consent form with explanation of the project, prior to the assessment. Only children whose parents agreed on participation (informed consent) participated in the study. All parents agreed on participation.

2.2. Data collection procedure

To answer the research question on language disturbances and their underlying neurolinguistic processes the production of the following elements was compared for the 3 groups of participants:

(a) percentage of primary plot elements, secondary plot elements or noise;
(b) percentage of sentence structures produced correctly at first or second attempt;
(c) percentage of incomplete or ungrammatical sentences and

(d) the type and frequency of disfluencies in retelling a memorized story.

The PCI contains 33 symptoms associated with cluttering, in four domains (pragmatics, speech motor, language and cognition & motor coordination and writing problems). Every symptom can be ranked with a score on a seven-point scale (0 = not present, 6 = always present) in order to predict possible cluttering.

Six researchers (four student researchers and two SLPs specialized in stuttering) were responsible for data collection and data-analysis. Data collection was, according to a specially developed protocol, achieved in the school of the children by bachelor students of Fontys University of Applied Sciences in Eindhoven, the Netherlands. All data was digitally recorded using PRAAT (Weenink & Boersma, 2008) software and coded by the first author. The main researcher was also responsible for coordinating data collection.

Recordings were made in a sound protected room. Digital audio- and video- tape recordings were made of all speaking tasks using a Sony digital video camera, a Trust digital head microphone, and a GoldWave Digital Audio Editor v5.18. All speech utterances produced by the individual participants were recorded through a Trust head microphone into a high quality sound card using a Compaq 6710b laptop, sample frequency 22.050 Hz. All spoken text was transcribed on the score form (see appendix). The number of primary plot elements, secondary plot elements and noise in retelling the story was counted and transferred to form B. All sentences were judged on syntactic correctness. The percentage and type of disfluencies was determined. In assessing sentence structures (like is described above) distinctions are made between grammatically correct sentence structures that were produced directly (before auditory monitoring) and after auditory monitoring (indirect). The analysis protocol thoroughly described both data analysis procedure and decisions concerning error analysis.

Material

The Wallet story (van Zaalen & Bochane, 2007), based on Renfrew's (1997) Bus story, was used in retelling a memorized story. In this standardised story, the primary and secondary plot elements are determined in advance and described in an assessment and analysis protocol (see Appendix). The researcher read the story out loud. After reading aloud, the child received a hand sign from the examiner to retell the story without help of the researcher. When the child was certain about the procedure the researcher started to read. Digital audio recordings were made.

2.3 Data analysis

Data analysis was undertaken by five researchers (four student researchers and one SLP) and blinded for diagnostic group. Prior to data analysis the students and the SLP received a two day course in analysis of the narrative and syntactic elements of the narrative by the principal investigator. Data-analysis consisted of determining:

a) The percentage of primary plot elements, secondary plot elements and noise used in retelling a memorized story. Primary plot elements are defined as 'the building stones of the story' and secondary plot elements as 'details'. Noise is defined as added phrases not being part of the story (for instance, "I do not remember the story").

b) The percentage of grammatically correct sentence structures at first attempt (CSSf). A sentence is counted as a CSSF, when a correct sentence structure is produced immediately or after reformulating in reaction on internal monitoring. In order to do this, sentence structures were judged after deletion of word and phrase repetitions.

c) The percentage of grammatically correct sentence structures at second attempt (CSSs). A sentence is counted as an ICSS when a grammatically correct sentence structure is produced after reassembling sentence structure in reaction to auditory monitoring of discovered inadequacies. In order to do this, sentence structures were judged again after deletion of revisions and false starts.

d) The percentage of grammatical sentence structure errors (SSE). A sentence is counted as an SSE, when after both internal and external monitoring still syntactic, semantic or morphological errors are produced. In order to do this, sentence structures were judged after deletion of word- and phrase repetitions, revisions and false starts.

e) Ratio Disfluencies (RDF) and type of normal disfluencies. The frequency and type of disfluencies is determined. Accordingly the amount of normal disfluencies compared to the amount of stutter like disfluencies, the RDF, is determined (see vanZaalen et al., 2008a). Disfluencies were mainly categorised according to Campbell and Hill's Systematic Dysfluency Analysis (1987). Contrary to Campbell and Hill, word repetitions produced without extra tension were scored as normal disfluencies, tensed word repetitions were scored as stutter-like disfluency.

f) Total score on the Checklist Broddel Kenmerken (van Zaalen, 2008).

2.4. Statistical analysis

To compare the underlying language production processes of CWC or LD-children, language production in retelling a memorized story was compared to mean results in the control group. Analysis of variance between diagnostic groups is studied for statistical difference ($p < .01$). Statistical analysis, using SPSS 16.0, was controlled for difference in group size using Tukey-s b procedure.

Reliability

In order to obtain reliable data, children who produced less than 100 words in retelling the Wallet story were excluded from this study. Reliability and validity of data is of essential importance, therefore inter- and intra- judge reliability scores of multiple analyses were determined. To establish inter- and intra- judge reliability for the Sentence Structure scores, three written out stories were judged (without information of diagnostic group) by all researchers on two different occasions. After that, mutual correlation was counted. Reliability was considered satisfactory when inter judge reliability correlation scores of 0.80 were reached.

3. RESULTS

Four LD-children did not meet the 100words criterion and were excluded from the study. Satisfactory correlations for inter- judge reliability scores for story issues ($r = .980$), sentence structure correctness ($r = .881$), type of disfluencies ($r = .997$), and RDF ($r = .974$) were reached.

	Cluttering (N=11)	Learning disabled (N=33)	Controls (N=55)	*F*	*P*
PPE	9.2 (1.8)	5.97 (2.04)	9.1 (1.4)	38.294	.0001
SPE	3.4 (1.7)	2.8 (1.6)	4.4 (1.1)	15.662	.0001
Noise	0.8 (0.8)	1.4 (1.4)	1.8 (1.5)	2.697	.072
CSSf	72.8 (19.4)	57.4 (17.1)	83.5 (7.5)	41.812	.0001
CSSs	88.7 (7.1)	80.0 (14.6)	91.8 (6.0)	15.252	.0001
SSE	11.6 (7.2)	20.2 (14.5)	8.1 (6.0)	16.296	.0001
RD	8.4 (78.5)	13.0 (87.6)	8.1 (47.8)	5.509	.006
NDF	12.8 (5.7)	15.1 (13.1)	7.7 (5.0)	8.552	.0001
PCI	66.6	61.5	38.9	9.327	.0001

Table 1.: Language production characteristics and analyses of variance in CWC, have learning disability compared to controls. Abbr.: PPE: primary plot element; SPE: secondary plot element; CSSf: correct sentence structure first attempt; CSSs: correct sentence structure second attempt; SSE: sentence structure errors; RD: ratio disfluencies; NDF: percentage normal disfluencies.

A) Primary plot elements, secondary plot elements and noise

Between group analysis of variance showed that the percentages of primary plot elements and secondary plot elements were statistically significant between groups (primary plot elements [F=(2,98)=38.294 ; p<.00001]; secondary plot elements [F=(2,98)=15.662, p < .0001]; No difference was found for noise elements [F(2,98)=2.697, *p*= .072]. LD-children produced fewer primary plot elements in retelling a memorized story compared to controls and CWC (LD: *M*=6.0, *SD*=1.4; CWC:*M*=9.2, *SD*=1.8; controls: *M*=9.1 *SD*=1.4). Controls reproduced more secondary plot elements compared to LD-children (controls: *M*=4.4, *SD*=1.1; LD: *M*= 2.8, *SD*=1.6; CWC: *M*=3.4, *SD*=1.4) (see table 1 & figure 2).

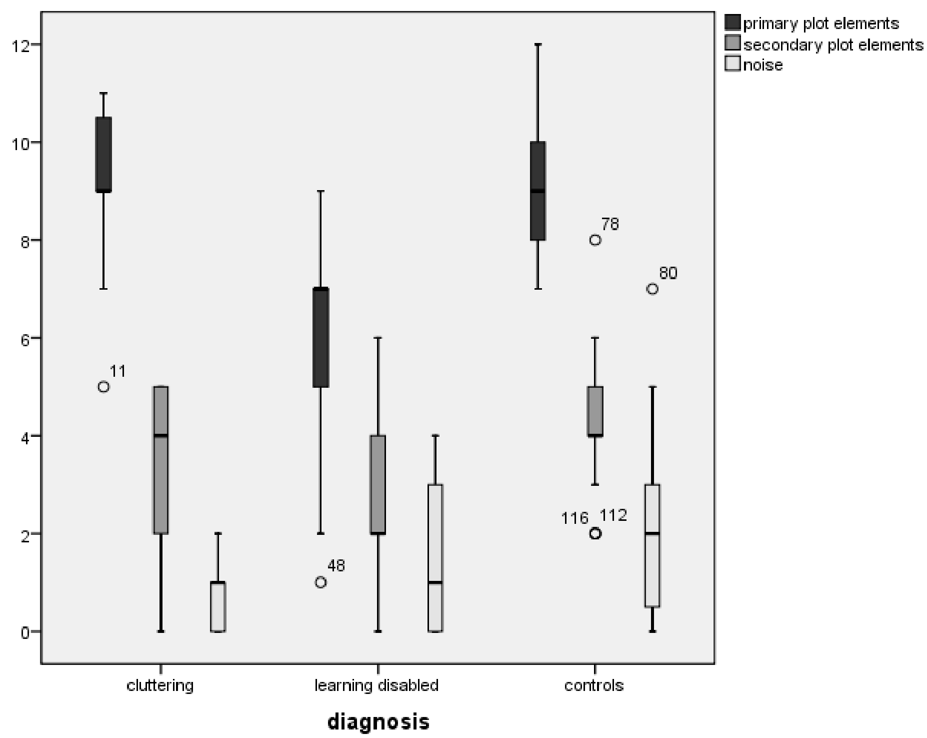

Figure 2. Number of reproduced story components per diagnostic group.

B) Sentence structures

Multiple group comparison showed that there was a statistically significant difference between groups on production of grammatical correct sentence structures at the first attempt (CSSf), [F(2,98) = 41.812; p < .0001], production of correct sentence structures at second attempt (CSSs), [F(2,98)= 15.252; p< .0001]. LD-children produced fewer correct sentence structures at first attempt compared to CWC or controls (CSSf LD: *M*=57.41%, *SD*=17.1; CWC: *M*=72.82%, *SD*=19.4; controls: *M*=83.54%, *SD*=7.46), (see Table 1).

LD-children produced fewer correct sentence structures at the second attempt compared to CWC or controls (percentage CSSs LD: *M*=80.03, *SD*=14.61; CWC: *M*=88.73%, *SD*=7.03; controls: *M*=91.82%, *SD*=6.04), (see Table 1).

C) Sentence Structures Errors (SSE)

A between group analysis of variance showed that there was a statistically significant difference between groups on the production of grammatical incorrect sentence structures [F(2,98)=16.432, p< .0001]. LD-children produced significantly more incorrect sentence structures compared to controls and CWC. (LD: *M*= 20,2%, SD=14.6; CWC: *M*=11,4%, *SD*= 7.2; Controls: *M*=8.1, *SD*=6.0), (see Table 1 & Figure 3).

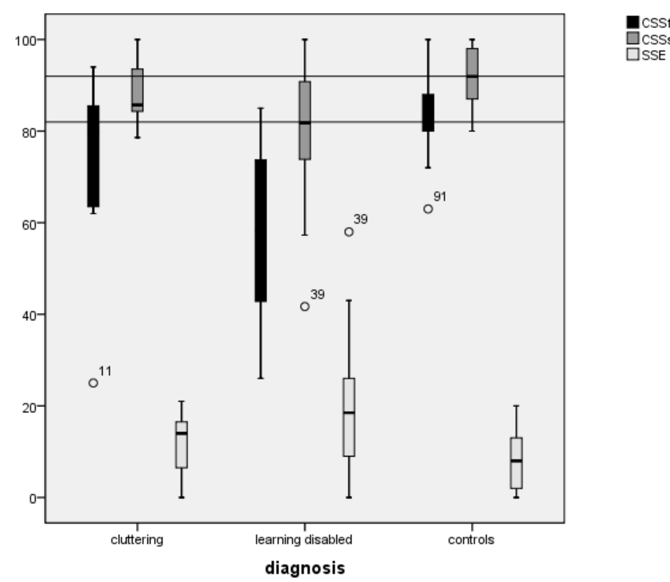

Figure 3. Results on sentence structure production in percentages per diagnostic group on correct sentence structures in first attempt (CSSf), correct sentence structures on second attempt (CSSs) and sentence structure errors (SSE); (black line: mean CSSf in controls; grey line: mean CSSs in controls).

D) Ratio disfluencies

Analysis of variance on ratio disfluencies (RDF) showed there was a statistically significant probability of a difference between groups in RDF in retelling a memorized story [F(2,98) = 5.509; *p*= .006]. Although LD-children produced a much higher amount of disfluencies compared to CWC and controls (LD: *M*=13.0, *SD*=6.8; CWC: *M*=8.4, *SD*=7.9; Controls: *M*=8.1, *SD*=4.8) the probability of a difference between groups on RDF was not statistically different on the p < .05 level after Post hoc Tukey's b correction for unequal group size (see Table 2).

E) Type of normal disfluencies

Between group analysis of variance on types of normal disfluencies showed that there was a statistically significant probability of a difference between groups for word- and phrase repetitions [F(2,98)= 26.094, p< .0001]. CWC produced significantly more word- and phrase repetitions compared to LD-children and controls (CWC: *M*= 5.82, *SD*= 3.2; LD: *M*=2.01, *SD*=2.4; controls: *M*= .99, *SD*= 1.47).

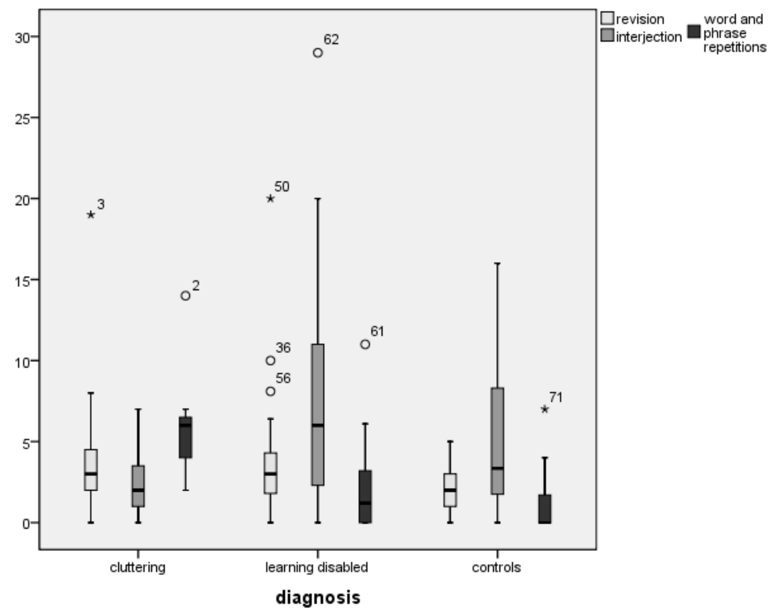

Figure 4.: Distribution (including outliers) of normal disfluencies: revision, interjection and word/phrase repetitions per diagnostic group

The probability of a difference on the production of uh-interjections was statistically significant between LD-children and other children (F(2,98)= 6.143; p = .003). Between group analysis of variance showed that there was a statistically significant probability of a difference between groups on revisions [F(2,98)=5.569, p= .005]. The probability of a difference on revisions between CWC and LD-children compared to controls was statistically different on the p < .05 level after Post hoc Tukey's b correction for unequal group size (controls: *M*= 1.97, *SD*= 1.40; CWC: *M*= 4.55, *SD*= 5.3; LD: *M*= 3.62, *SD*= 3.69). The statistical significant probability was not at the level of chance (see Figure 4).

F) Predictive Cluttering Inventory - Dutch revised

Scores on the Predictive Cluttering inventory – Dutch revised (CBK) were compared between groups and revealed significant group differences F (2,90)= 9.327, *p*< .0001). Controls scored a lower mean CBK score (38.94) compared to learning disabled (61.48) and CWC (75.58), these differences were significantly different (*p*=.002 for LD: *p* < .0001 for CWC). Differences between cluttering and learning disabled children were not significantly different (*p*= 1.000), see Table 1. For different reasons, the CBK list of eight LD-children was not administered by the parents (“forgotten” (5x) or “too difficult”(3x)).

4. DISCUSSION

The focus of this study is to ascertain whether disturbances in the fluency of language production of CWC can be linked to difficulties in the same underlying processes of language formulation in LD-children. Results indicated that language disturbances of CWC can be based on other underlying processes (conceptualizing and formulating) compared to those of LD-children.

Children with cluttered speech

Children with cluttered speech produced primary plot elements and secondary plot elements according to the normal level. Different authors (Daly & Cantrell, 2006; St. Louis, 1992; St. Louis et al., 2007; Teigland, 1996; Ward, 2006; Weiss, 1964) reported that PWC experience problems in organizing linguistic information. In this study, CWC did not experience story organization problems. This finding may be attributed to the fact that the speech task restricted the options available to the speaker and provided a story organization.

Speakers need to gain extra time for planning whenever difficult material (e.g., a content word or sentence structure) is not ready. They can do this by pausing or fluently repeating one or more prior segments (Howell & Dworzynski, 2005). After deletion of the word repetitions and revisions in CWC syntactically correct sentences appeared, comparable to controls. In producing repetitions of already produced words CWC seem to gain time for their formulation process (Ward, 2006). CWC seem to use the editing phase of the formulating process to produce grammatically correct sentence structures.

From the results in this study it can be concluded that language disturbances in CWC are mainly based on problems in the formulator (correct sentence structures after removing normal disfluencies), that disappear when a child who clutters has enough time to structure the sentences in a grammatical correct way. According to Ward (2006) CWC may experience problems at all three levels of speech production i.e. conceptualization, formulation and articulation. In this assessment the function of the articulator was left out. Problems in the conceptualiser (organising the story) were not expected in the used speech task. In retelling a memorized story read aloud by the researcher the concepts of the story and the linearization were restricted. It can be hypothesised that CWC produced sentence revisions in an attempt to imitate the example sentences as accurate as possible. Further research is needed to confirm the role of conceptualiser in cluttering

It is speculated that language production disturbances from CWC are mainly caused by a speech rate which exceed the limits of their retrieval skills, and that this results in insufficient time to allow adequate sentence construction. In a delayed reaction to their internal monitor or by auditory monitoring, CWC use sentence revisions and repetitions to help produce a correct sentence structure (Ward, 2006, p.145). This hypothesis is supported by the clinical observation that CWC are able to produce correct sentence structure in writing (St. Louis & Cantrell, 2006; Ward, 2006). Further research on language production at different rate levels is needed to confirm this hypothesis.

Children with learning disability

LD-children produced less primary and secondary plot elements compared to controls and CWC. The percentage correct sentence structures at first attempt by LD-children was significantly lower compared to controls and CWC. Disfluencies of LD-children were mainly characterised by within-sentence uh-interjections and revisions. The fluency of the LD-children was probably negatively influenced by problems in both the conceptualiser and the formulator. LD-children appear to have difficulties in retrieving the necessary lemmas within the communication time frame (Messer & Dockrell, 2006). In contrast to CWC, a high percentage of syntactical errors remained in the language production of LD-children, after deletion of the normal disfluencies. These children seem to be not able to fully complete the editing phase. The errors in syntactic structures do not simply imply that the person’s syntactic skills are weakly developed. Firstly, it can be a sign of significant problems in the conceptualiser (thought organising weakens the attention to sentence structure). Secondly, the LD-children can experience problems in selecting and ordering relevant information due to weak storage of word forms or a small lexicon (German, 1979, 1984). This might be especially true when the children also have to consider what has previously been stated by the researcher. Thirdly, word retrieval could be achieved inaccurately, more slowly or it may fail altogether and syntax errors are caused by incorrect lemma retrieval (German, 1979, 1984, 1992). It is hypothesized that the language disturbances of LD-children are mainly a result of problems in lexicon and lemma retrieval resulting in a high level of uh-interjections (assuming that these interjections indicate word-finding problems) and sentence structure errors.

LD-children also experience omission or substitutions of syntax units in writing. LD-children cannot use words to express organized and complete thoughts in writing. According to Graham and Harris (2002) it is not unusual for a child with writing and learning difficulties, to experience challenges generating content, executing the mechanical aspects of writing, and planning in advance, preferring to get ideas while writing. Furthermore, according to Graham and Harris, LD-children appear to use a least-effort strategy when revising, as the changes they initiate are primarily limited to minor word substitutions and unsuccessful attempts to correct errors of spelling, punctuation, and capitalization (Graham & Harris, 2002). So, it can be concluded that the conceptualisation and formulation problems of LD-children are not limited to spoken language alone.

Language production in retelling a story

In this study, language production was controlled by the speech task: retelling a memorized story. For instance in retelling a story, the example utterance restricts the options available to the speaker concerning the way he formulates a repair (Levelt, 1992). The functional significance of this is clear: the transfer of these sorts of restrictions, from one utterance to the next, will at the same time increase the fluency of speech (by reducing the number or size of the formulator's operations), and the coherence of discourse (by establishing structural relations between present and previous speech) (Levelt, 1983).

The value of Renfrew's bus-story as a predictor of language development has been demonstrated by its use in follow-up studies of children with disabilities in spoken language (Bishop & Edmundson, 1987). Assessment of language production in CWC or LD-children was not performed earlier. Knowledge on language production was mainly based on clinical observations (Ward, 2006). Although the Wallet-story, used in this research, differentiated language production disturbances of CWC or LD-children, further research on the Wallet story is needed, to provide norms on the Wallet-story in the population of older children, adolescents and adults. In this study sentence structure complexity was not examined. Further research on complexity of sentence structure can provide more detailed information on syntactical abilities of both CWC and LD-children.

Disfluency Ratio

Language production has separate planning and execution components (Levelt, 1989). Speakers can start an utterance before they have the complete plan. Covert and overt repairs are responses to detected speech errors or inappropriateness, heard as disfluencies (Kolk & Postma, 1997). Both CWC and those who have learning disabilities produced a high frequency of normal disfluencies. The ratio of non-stutter like disfluencies and stutter like disfluencies (RD= NSD/SD) of LD-children is higher than in CWC or controls. This can be explained by the fact that in contrast to LD-children, CWC also produced a small number of part-word repetitions that, according to Campbell and Hill (1987) belong to stutter like disfluencies. One explanation for this is that CWC have better functioning internal monitors than LD-children.

Predictive Cluttering Inventory

Results of the Predictive Cluttering Inventory showed no significant differences between CWC and LD-children. One explanation is that cluttering and learning disabilities have many characteristics in common, as is stated by many researchers (Daly & St. Louis, 1986; Mensink-Ypma, 1990; St. Louis, 1992; St. Louis, Myers, Bakker & Raphael, 2007 in Curlee & Conture, 2007; Tiger, Irvine & Reis, 1980; Ward, 2006; Weiss, 1964). Another explanation can be that the low level of interjudge reliability scores influenced on the sensitivity and specificity of the checklist. In order to use this checklist to detect possible cluttering elements one should notice that further research on the instructions, specificity and the sensitivity of this screening instrument is needed.

Clinical implications

This research produced some insights on differences and similarities in underlying processes of language disturbances in CWC or LD-children while retelling a memorized story. In planning therapy for fluent LD-children, our results suggest that these children are best supported with training of all aspects of conceptualisation and formulation. In planning therapy for CWC our results indicate that adjustment of communicative rate of the children and their environment should have first priority, in order to gain time for adequate formulation.

Conclusion

Results of this study confirm the differences in underlying processes of language disturbances between children with cluttered speech and LD-children. While language production of LD-children was disturbed by problems in conceptualiser and formulator, language planning disturbances in CWC were considered to arise due from insufficient time to complete the editing phase of sentence structuring. Results indicate that CWC can be differentiated from LD-children by both the number of main and secondary plot elements and by the percentage correct sentence structures.

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Appendix 1

"The Wallet Story": retelling a memorized story

Instructions for task administors:

"I will read you a story.

After I have finished reading the story, you will retell the same story as precisely as possible to me.

I am not allowed to help you with it.

When I am ready, I will give you a sign.

Then it is your turn to tell me the same story". If the child indicates that he/she has understood the instructions, do start to read the story. If the child has not understood, give further explanations, as required, until the child understands the procedure.

Digital audio recording starts as soon as the therapist starts to read.

Analysis: (use score form of appendix)

1. Transcribe the story told by the client.
2. Determine story components (main issues, side issues and noise).
3. Determine syntax correctness (+ = correct; - = false).
4. Determine type of disfluencies.
5. Determine percentage disfluencies and ratio disfluencies.

Score form: The wallet story

Therapist:

Client's name:

Birth Date:

Main issues	**Side issues**	**Story**	**Client's response**	**Syntax (not) correct**	**Dysfluency**
1		It was a rainy day in November			
2		A woman drove to the hypermarket in her brand-new car.			
3		She invited three girlfriends to dinner tonight			
	1	and had promised them to prepare something Italian			
	2	What is her speciality?			
4		While she was cooking the dinner			
5		her wallet fell from its case			
6		but she did not notice it			
	3	Her trolley was already loaded			
7		When she arrived at the pay-desk she could not pay for her groceries.			
	4	The cashier was willing to watch her caddie for a while			
	5	The woman put her groceries aside			
8		and went home			
	6	The square wipers of the car fell quickly up and down			
	7	And all traffic lights she encountered were of course red			
	8	She was terribly fed up!			
9		Just when she opened her house door			
10		the phone started ringing			
11		A little boy told her			
12		he had found her wallet			
	9	The woman was very relieved			
13		That was the story			

Story Issues (almost) fully mentioned**:**	**Normal disfluencies:**	**Stutter like disfluencies:**
... Main issues	- Revision	- Word repetition (tensed)
... Side issues	- Interjection	- Part word repetition
… Additional issues (= noise)	- Word repetition (no tension)	- Prolongation
	- Phrase repetition	- Block

PART two

Underlying neurolinguistic processes

Chapter SIX

Cluttering & Stuttering : different disorders.

A neuro-imaging study

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submitted

Abstract

The purpose of this study is to see if there are differences in brain activity involved in motor control and coordination during the performance of a range of spoken tasks (increasing in linguistic or motor complexity) in adults who clutter (AWC) compared to adults who stutter (AWS). fMRI data was collected in a Philips 3T scanner from a group of 13 right handed adults who stutter but did not clutter (AWS) and 13 right handed adults who clutter but did not stutter (AWC). The tasks involved speaking words of increasing complexity. Utilizing a block design with a 40 second on-off period that alternated viewing symbols with reading and speaking words was used.

Both groups showed speech related BOLD activity predominantly in the frontal lobe and sub-cortical nuclei. Group contrasts indicated that AWC showed higher activity in right hemisphere precentral gyrus, inferior frontal gyrus and left insula. AWS showed higher activation in right primary motor cortex, temporal lobe and globus pallidus. Findings tentatively suggest deficits in different parts of the brain are responsible for cluttering and stuttering.

Introduction

Cluttering is a disorder which has proved hard to define succinctly. The core symptoms of cluttering are a fast speech rate and jerky speech rhythm (St Louis, Myers, Raphael & Bakker, 2007). PWC can also show a high frequency of normal disfluencies (revisions, interjections, repetition of words or phrases), problems in placement of pausing in sentences or syllable structure errors (especially in multisyllabic complex words), (Daly & Cantrell, 2006; Hartinger & Mooshammer, 2005; St. Louis, Hinzman & Hull, 1985; VanZaalen, Wijnen & Dejonckere, 2009a; Ward, 2006). When PWC do not try to monitor their speech this tends to result in increased disfluency (St. Louis et al., 2007; VanZaalen & Winkelman, 2009; Ward, 2006; Weiss, 1964), while increased alertness commonly suppresses the cluttering characteristics. These characteristics together make the speech of AWC disorganized and sometimes difficult to comprehend (Daly & Burnett, 1996; Damsté, 1984; Myers, St. Louis, 1996; St. Louis et al., 2003; Van Zaalen & Winkelman, 2009).

Significant in regard to the diagnosis of cluttering is its relationship with stuttering. It is known that these two disorders commonly co-occur. Some authors even have claimed:

"*that stuttering has his roots in cluttering*" (Weiss, 1964, p.5). At present, the relationship between stuttering and cluttering is unclear. A major point in this study is to clarify the notion of 'cluttering' as nosologically different from stuttering.

Adults who stutter (AWS) and adults who clutter (AWC) have nonfluent speech in common, however, there are some important differences in speech output. In contrast to cluttering, the speech of adults who stutter is, according to Guitar (1998) characterized by: "*an abnormally high frequency or duration of blocks in speech flow"* (Guitar, 1998, p10). In addition, there may also be tensed repetition of sounds, syllables or words and prolongation of sounds (Craig et al., 1996; Craig, 2000; Peters & Guitar, 1991; Shapiro, 1998), whilst many PWS also acquire secondary behaviours which may function as strategies to reduce fear of prolongations and repetitions (Bakker, 1996; Craig, 2000; Roodvoets, 1987).

In stuttering, structural linguistic factors influence both the location and frequency of the fluency disruptions, for example word class (Howell & Au Yeung,2002), position of a word within a sentence (Howell & Au Yeung, 1995). Utterance length also has an effect on stuttering frequency (Bernstein-Ratner, 1997). Cluttering too tends to occur under increased linguistic demand, for example in longer utterances, and when more complex syntactic and semantic processing is required (Ward, 2006). But although both stuttering and cluttering seem to be affected by language variables, the manner in which this happens appears to be different. While in stuttering, linguistic complexity at various levels often leads to an increase of stutter-like disfluencies, in cluttering linguistic complexity tends to lead to an increase of speech errors and repairs (VanZaalen et al., 2009a, b).

Brain activation during reading in stuttering

There is now a considerable data base on brain functioning in stuttering (e.g. Guitar, 2006; Ward, 2006 for recent reviews). Recent fMRI research (Viswanath et al., 2003) showed increased Blood-Oxygen-Level Dependent (BOLD) response in PWS during reading in speech motor centres in the non-dominant hemisphere. These results were confirmed by several other researchers (Fox, 1996; Fox, 2000; Ingham, 2000; Neumann et al., 2003; Preibisch et al., 2003). Viswanath, et al., (2003) also observed reduced activity in the gyrus precentralis (both R and L) and gyrus post centralis (left hemisphere) in word reading of AWS. Neumann et al., (2003) and Preibisch et al., (2003) also observed increased activation in non dominant precentral sensorimotor and frontal regions in 9 right-handed adults who stuttered. When studied at follow up, after therapy, activity in this region (of the Inferior Frontal Gyrus) had significantly decreased (Neuman et al., 2003). Reduced activation of the left frontal precentral cortex was also found in children who stutter (CWS) by Watkins et al., (2008) at a location slightly posterior and dorsal to the peak of the ventral premotor cortex.

Because cluttering is hypothesized to be a language based fluency disorder affected by rate and stuttering a disorder of motor execution influenced by linguistic complexity, a dynamic paradigm with both speech motor and language components is needed in this study. In a speech elicitation paradigm with different levels of linguistic complexity, speech motor control can be examined at syllable and word level. Phonological and motor planning at syllable level ([pə], [təkə] and [pətəkə], (paradigm 1) is relatively easy. Nonword recall tasks are believed to reflect phonological working memory, since they must be carried out independent of semantic lexical knowledge (Gathercole & Baddeley, 1990). Adult speakers are able to repeatedly produce and recall programmed syllables (Riley & Riley, 1985). In producing multisyllabic word strings subjects experience challenges in both phonological planning and speech motor execution. Lexical selection is sensitive to word frequency on the level at which the phonological properties of words are retrieved (Navarette, Basagni, Alario, Costa, 2006). In frequently used multisyllabic word strings (paradigm 2) planning complex sequences of movements will be done faster compared to low frequency word strings (Baddeley, Gathercole & Papagno, 1998). In low frequency multisyllabic strings of words (paradigm 3) phonological planning is rather complex because novel phonological forms of new words take longer retrieval time.

In this paper we consider pure cluttering and pure stuttering as separate and distinct fluency disorders with different pathogeneses. Cluttering is hypothesized to be a language based fluency disorder affected by speaking rate (Weiss, 1964; Daly, 1996; VanZaalen, Wijnen & Dejonckere, 2009c). The core dysfunction in stuttering suggested by Alm (2004) is the impaired ability of the basal ganglia to produce timing cues for the initiation of the next motor segment in speech (Alm, 2004). If cluttering and stuttering are two independent disorders of fluency with different pathogeneses, it is reasonable to hypothesise that adults who clutter or stutter display different neurocognitive processes when performing speech tasks that call upon increasing demands on speech motor and linguistic skills. However, in comparing two disorder groups only relative differences between groups can be studied.

We therefore assume that PWC will show speech related BOLD activity predominantly in the frontal lobe and sub-cortical nuclei, as is known for stuttering and fluent speakers.

Basal ganglia circuits are associated with a variety of functions including motor control and learning. Basal ganglia dysfunction is implicated in disorders of movement and behavior control such as Huntington disease, Tourette's syndrome, obsessive compulsive disorder, Parkinson's disease and stuttering (Alm, 2004, 2006). When stuttering and cluttering indeed are disorders of speech fluency in which neurocognitive processes are disturbed in a different way a difference in activation levels of basal ganglia circuits can be expected in adults who clutter and those who stutter. Especially, while behavioural inhibition problems as impulsiveness, distractibility and excessive speed of delivery, are well known in cluttering, contrary to stuttering (Weiss, 1964).

Brain activation is higher when motor execution demands are higher. As AWS experience significant problems in speech motor execution, a higher activation level in primary motor BOLD responses can be expected as AWS have to perform speech tasks which complex phonological and motor planning (see paradigm 3). The time course of lexical activation and selection, phonological encoding, and phonetic encoding is assumed to be lower in short and high frequency words. As a result of this activation levels in primary motor cortex in multisyllabic high frequency word strings (paradigm 2) or syllables (paradigm 1) are assumed to be relatively lower compared to paradigm 3.

Increased activation in right hemisphere (RH) precentral sensorimotor and frontal regions was seen in adults who stutter before effective fluency therapy (Preibisch et al., 2003). This increased right hemisphere activation was interpreted as a compensation to regain fluency (Neuman et al., 2003; Preibisch et al., 2003). As cluttering is hypothesised to be a language based fluency disorder affected by rate, it is expected that AWC will experience phonological planning problems in producing word strings at a fast rate. The ventral premotor cortex is involved in planning complex sequences of movements (Watkins et al., 2008). As AWC produce significantly more errors in planning complex sequences of syllables compared to AWS (van Zaalen et al., 2009a) a difference in ventral premotor BOLD responses can be expected when AWC and AWS have to perform speech tasks in which they have to phonologically encode and produce complex syllable strings. As a result of these problems we assume that higher relative BOLD activity in language planning areas of the non-dominant hemisphere will be seen in adults who clutter compared to AWS, as a compensatory strategy to regain linguistic fluency.

Motion artifacts

Artifacts in overt speaking during fMRI scanning can be caused by bulk head motion (Bullmore et al., 1999), which could potentially be corrected for (Friston et al., 1995a; Hajnal et al., 1995; Woods et al., 1993) or diminished by wearing a stabilising helmet.

Another cause of artifacts during overt speaking can be by magnetic field variations caused by the changing pharyngeal space during speaking (Birn et al., 1998) leading to signal fluctuations and image distortions that depend on the individual anatomy (Preibisch, Raab, Neumann, Euler, von Gudenberg, Gall, 2003). Therefore, in dysfluent persons it would be desirable to examine more extended speaking periods, because dysfluency symptoms occur more often in longer and grammatically complex utterances than in shorter and simpler ones (Wingate, 1988; Anderson & Conture, 2000). Prolonged stimulus durations and repetition times of 3 seconds will allow an effective suppression of speech-related artifacts in fluent as well as in nonfluent speakers (Preibisch et al., 2003). The experimental design should at least permit the production of repeated syllable strings with a duration of 5 seconds to comply with this duration.

Although, Ingham et al., (2000) found evidence that motor speech centres are affected even during imaginary stuttering (Ward, 2006, p.32) using imaginary cluttering is no option. Cluttering happens while people are unaware of mistakes and have unvalid focus to speech production. Imitation of cluttered speech is only possible with focus on speech production. In order to diminish motion artifacts participants should be trained to speak with minimum tongue, jaw and lip movement and minimal voicing. In performing this '*whispered speech*' possible artifactual effects are limited because of this and supposed to be equal for all participants.

Furthermore, it is important in fMRI research in fluency disorders to train the subjects in performing the speech task, before the subjects are placed inside the magnet. Training should contain instructions to repeat the target words seen on the screen at their fastest rate, while still remaining intelligible. Repetition should start after the target is shown and remain till the next target is shown on the screen.

When indeed differences between cluttering and stuttering adults are found, further research including adults without fluency problems as a control group can identify basic functional differences.

Aim of the study

The difference in the behavioural profiles of AWC and AWS is expected to be reflected in these individuals' patterns of brain activity. The purpose of this study is to see if there are differences in brain activity involved in motor control and coordination during the performance of a range of spoken tasks (increasing in linguistic or motor complexity) in AWC compared to AWS. Specifically we wish to examine whether the activity level of the ventral premotor cortex (of the non-dominant hemisphere), basal ganglia centres and primary motor cortex is different in AWC compared to AWS.

2. METHOD

2.1 Participants

Participants were thirty healthy, right handed individuals ranging in age from 18.3 to 47.5 years. Stuttering (*N*=16) and cluttering subjects (*N*=14) were recruited from a group of people who joined a self help group for disfluent persons. In this study no controls were recruited, because the aim of the study was to detect whether brain activation of 'pure' PWC was different from 'pure' stutterers. Participants were randomly selected volunteers diagnosed with either pure cluttering or pure stuttering from this opportunity sample. Case history of all subjects revealed no concomitant disorders diagnosed (no history of ADHD or ASD).

2.1.2 Diagnostic criteria

Participants were diagnosed with cluttering or stuttering based on internationally accepted perceptual characteristics of these disorders described by among others Daly (2008), St. Louis et al., (2003, 2007) and VanZaalen et al., (2009c):

Cluttering diagnosis

Persons were diagnosed with pure cluttering and included in the study when they fit all of the following four criteria:

(1) a fast and/or irregular articulatory rate was observed, combined with:
(a) errors in syllable or word structure; or
(b) a high frequency normal disfluencies and/or
(c) extra (non-linguistic) pausing;
(2) SSI-3 severity scores were equal or below 2 (no – mild stuttering);

(3) the person had no coincidental language problem: the person was able to produce correct sentence structures without semantic or morphological errors in both spontaneous and written speech.
(4) the score on the Brutten speech situation checklist (S-24) was within normal limits.

Stuttering diagnosis
Persons were diagnosed as pure stuttering and included in the study when they fit all of the following five criteria:
(1) prolongations and or blocks longer than 0.5 seconds were determined (Guitar, 2006);
(2) the ratio disfluencies was < 1.0;
(3) Stuttering severity, SSI-3 scores, was at least moderate.
(4) subjects did not experience errors in syllable or word structure and pausing.
(5) the person had no coincidental language problem: the person was able to produce correct sentence structures without semantic or morphological errors in both spontaneous and written speech.

All participants spoke Dutch as their first language and all had a diploma on higher education or university level.

<u>Excluding criteria</u>
a) PWC presenting with language disorders were not considered pure clutterers. In order to analyse language production a transcript of spontaneous speech was analysed and sentence structure was observed after removal of normal disfluencies. For instance, the sentence structure in: "*I (get..,) getting on my bike*", was considered to be syntactically incorrect. If the AWC was not able to compile syntactically correct sentence structures, in spontaneous speech, or written language they were excluded from the study.
b) PWS who showed speech related dyskinesias which may have resulted in movement artifacts were excluded from the study.
c) Participants with a weight over 130 Kgs, with claustrophobia, dyslexia or implanted medical devices (e.g. pacemakers) were excluded from the study, as were people carying metal parts in their body.
d) Participants with less than 100% right handedness, as rated on the Edinburgh Handedness Inventory (Oldfield, 1971) were excluded from the study.

Subjects were informed about the scanning procedures, according to ethical rules of scientific research with people. Subjects gave their informed consent to be a volunteer, twice (with two weeks interval). The medical ethical committee of the Academic Medical Centre in Amsterdam approved the study (reference code: MEC 08/210 #08.17.1910). Assessment of speech and language characteristics took place before scanning. Participants were trained in performing the speech tasks (see task requirements section) before taking part in the experimental tasks in the MRI scanner.

2.1.3. Speech tasks for diagnosing stuttering and cluttering

Data were gathered on: articulatory rate, accuracy and smooth-flow, frequency and type of normal disfluencies, sentence production in writing and cognitive and emotional responses to the fluency disorder. The test sequence for all participants was: 1) monologue; 2) reading; 3) speech motor control at word level; 4) writing; 5) Speech situation checklist: S-24. For monologue, participants were asked to recount an event in the recent past of their own choosing without intervention of the speech therapist. Recordings lasted 3 - 5 minutes. For reading, participants were asked to read a standardised story (TVSE-L, Boey, 2000). In order to examine their skills on speech motor control on word level was tested using the Screening Pittige Articulatie (SPA, van Zaalen, Wijnen & Dejonckere, 2009a). SPA is an OMAS (Riley & Riley, 1985) based instrument that evaluates oral motor coordination at word level. Subjects were asked to repeat longer multisyllabic words at a fast rate. Accuracy, smooth-flow and rate were determined and compared to normative data (VanZaalen et al., 2009a). Writing ability was assessed by having subjects copy a written standard text and to write down a personal experience in the recent past (20 sentences). Brutten’s Speech situation checklist (S-24) was used to control for negative feelings or cognitions towards speech situations (Brutten in Janssen, 1990).

Recordings were made in a soundproof room. Digital audio- and video tape recordings were made of all speaking tasks using a Sony digital video camera, a Trust digital head microphone, and a GoldWave Digital Audio Editor v5.18. Audio files were recorded through a Trust head microphone into a high quality sound card using a HP Pavilion zv6000 laptop, sample frequency 22.050 Hz.

2.2 Speech analyses

Articulatory rate was determined using a speech analysis program, PRAAT (Boersma & Weenink, 2007). Stuttering severity was determined using the Stuttering Severity

Instrument (SSI-3, Riley & Riley, 1994). Syllable and word structure was analysed with the SPA (Van Zaalen et al., 2009a). Ratio of disfluencies was determined after counting the number of normal disfluencies (non tensed word or phrase repetitions, revisions, interjections) and the number of stuttering disfluencies (sound, syllable or tensed word repetitions, prolongations and blocks). The ratio was determined by dividing the normal disfluencies by the stutter like disfluencies (Van Zaalen, Wijnen & Dejonckere, 2009a; Van Zaalen & Winkelman, 2009).

2.3. Functional magnetic resonance imaging (fMRI)

All BOLD fMRI imaging was performed on a 3 Tesla Philips MRI-scanner. The fMRI session consisted of two parts. First, anatomical imaging of the brain was carried out. A spoiled gradient echo structural T1-weighed scan of 170 sagittal slices was made for coregistration with the fMRI data (voxel size 1x1x1 mm). For fMRI, subsequent whole-head imaging was performed with an optimized echo-planar imaging (EPI) sequence: TR= 2500 ms, TE = 30 ms, flip angle= 90^o, slices= 35, slice thickness= 3 mm, matrix = 92 x 92, FOV= 230 mm. A total of 285 volumes was acquired in each of the subjects.

2.3.1. Task requirements

Three paradigms were used to test different levels of speech programming and language planning:

(1) Repeated nonsense syllables (1-3 syllables) with labial, dorsal and velar plosives ([pə], [tə] and [kə], respectively);

(2) Repeated high frequency short multisyllabic words (3 syllables each);

(3) Repeated low frequency multisyllabic word strings (6-14 syllables), (see Table 2).

The multisyllabic words were created using mostly the same initial, medial and final consonants that were used in the first paradigm (see Table 1). The same consonants were used in order to exclude effects of place and manner of articulation between the paradigms.

	Sign string	**Words** (in Dutch)
Experiment 1	$!#, (!@^$#, ?<@*^$)*#/	/Puh/, /Tuhkuh/, /Puhtuhkuh/
Experiment 2	$#*@!%#%^^?, (^@^%!?<	/patatkraam/, /katapult/, /peperbus/, /takkenbos/
Experiment 3	@&&%$!&*&?)(&%&&@! *(%^$@!#*&*?<>+%$#@!&*	/opperceremoniemeester/ /vergevorderde experimenten/

Table 1.: Sing strings and words used in paradigms

Participants were trained to ‘whisper’ the target words as fast as they could, while still remaining intelligible before they were placed inside the magnet. Headphones were worn to protect against scanner noise, and subjects also wore a Sense head coil in order to minimize artifacts due to head movement. Participants were instructed to breath and swallow naturally.

2.3.2 Block design

To filter the activity of the visual cortex the speech motor and language paradigm was mirrored by a visual paradigm. Using a 40 sec. on/off block design, that contrasted reading symbols (participants were instructed to 'read' the symbols from left to right repeatedly as long as it was on the screen) with reading and speaking. The subjects undertook three experiments with speech tasks or reading strings of signs. fMRI recordings started with recordings of reading, followed by recordings of pronouncing syllable strings. Within each 40 seconds paradigm, five recordings of each eight seconds were made in a randomized order (see Figure 1.). Series of eight seconds speaking words or reading signs were used to prevent a drift in the BOLD signal (Birn et al., 2004).

All the words and sign strings were presented in the MRI-scanner using Presentation 12.0 and dedicated fMRI stimulus presentation equipment. Presentation controlled all external devices and sent timing pulses to the scanner and made start of stimulus sequences on specific pulse/scan possible. All the movement artifacts associated with speech were incorporated into the BOLD response estimation. In minimalizing jaw and tongue movement and in stabilizing the head and jaw we tried to exclude as many movement artifacts as possible.

2.3.3 Data analysis

FMRI data were analyzed in the context of the general linear model, using epoch models (box-car regressors) convolved with a canonical hemodynamic response function to model responses to each speech condition. Contrast images containing parameter estimates were entered into a second-level (random effects) analysis. Main effects across groups and group interactions were analyzed with a one-sample t-test implemented in SPM5 with diagnosis as covariate. Main effects across groups are reported at a $P < 0.05$ corrected for multiple comparisons according to the False Discovery Rate (FDR) method (Genovese et al., 2002). Whole-brain group interactions are reported at $P < 0.001$, masked with the appropriate main effect at $P < 0.05$ and minimum cluster size of 10 voxels.

A total of three contrasts was studied:

(1) the activation difference of the brain between reading words and sign strings in the cluttering group during the three experiments;

(2) the activation difference of the brain between reading words and sign strings in the stuttering group during the three experiments;

(3) the activation difference of the brain between the cluttering group and the stuttering group during the three experiments;

Areas with significant activation were identified in Talairach coordinate space and linked with corresponding Brodmann areas (Talairach & Tournoux, 1988). Because this was the first study design in which results of AWC and AWS were compared, results were compared to known fMRI results in other research projects with stuttering individuals and controls.

3. RESULTS

3.1. Diagnostic results

For a description of participants correlation with diagnostic characteristics participants, see Table 2.

Nr.	*S-24*	*Rate> 5.4 or Variation > 3.3 SPS*	*SPA z-score < -1*	*> 5% NDF or pausing errors*	*RD > 2.8*	*0 < SSI-severity score < 2*	*Syntactically correct sentences*	Prolongations or blocks > 0.5 sec	RD < 1.0	SSI-severity score ≥ 3	% NDF < 3%	SPA z-score ≥ 0	Diagnosis
1	+	+	+	+	+	+	+	-	-	-	-	-	PWC
2	+	+	+	+	+	+	+	-	-	-	-	-	PWC
3	-	-	-	-	-	-	+	+	+	+	+	+	PWS
4	-	-	-	-	-	-	+	+	+	+	+	+	PWS
5	+	+	+	-	-	+	+	+	-	-	-	-	PWCS**
6	+	+	+	+	+	+	+	-	-	-	-	-	PWC
7	-	-	-	-	-	-	+	+	+	+	+	+	PWS
8	-	-	-	-	-	-	+	+	+	+	+	+	PWS
9	+	+	+	+	+	+	+	-	-	-	-	-	PWC
10	-	-	-	-	-	-	+	+	+	+	+	+	PWS
11	-	-	-	-	-	-	+	+	+	+	+	+	PWS
12	-	-	-	-	-	-	+	+	+	+	+	+	PWS
13	+	+	+	+	+	+	+	-	-	-	-	-	PWC
14	-	-	-	-	-	-	+	+	+	+	+	+	PWS
15	+	+	+	+	+	+	+	-	-	-	-	-	PWC
16	-	-	-	-	-	-	+	+	+	+	+	+	PWS
17	+	+	+	+	+	+	+	-	-	-	-	-	PWC
18	+	+	-	-	-	+	+	-	-	-	-	-	PWCS**
19	+	+	+	+	+	+	+	-	-	-	-	-	PWC
20	-	-	-	-	-	-	+	+	+	+	+	+	PWS
21	+	+	+	+	+	+	+	-	-	-	-	-	PWC
22	+	+	+	+	+	+	+	-	-	-	-	-	PWC
23	+	+	+	+	+	+	+	-	-	-	-	-	PWC
24	+	+	+	+	+	+	+	-	-	-	-	-	PWC
25	-	-	-	-	-	-	+	+	+	+	+	+	PWS*
26	+	+	+	+	+	+	+	-	-	-	-	-	PWC
27	+	+	+	+	+	+	+	-	-	-	-	-	PWC
28	-	-	-	-	-	-	+	+	+	+	+	+	PWS***
29	+	+	+	+	+	+	+	-	-	-	-	-	PWC
30	-	-	-	-	-	-	+	+	+	+	+	+	PWS

Table 2.: Participants characteristics; * excluded due to suspected dyslexia; ** excluded due to both cluttering and stuttering criteria prior to fmri assessment * excluded due to physical complaints (neck hernia).

3.2. FMRI

All subjects reported that they had been able to maintain whispering at a fast rate in reading loud the test words. Debriefing of the subjects also showed that all subjects were able to remain reading the signs and whispering the words in a fast rate. Transition from one panel to the next prompted an easy paradigm shift. Participants debriefing was consisted with observation of mouth movement during scanning procedure by the researchers.

3.2. Activation within groups

3.2.1. *The cluttering group* (see Table 3.)

Significant left cerebrum activation in AWC was located in primary motor cortex, premotor cortex and SMA in all paradigms. Furthermore, in paradigm 3, significant activation was located in left and right basal ganglia centres and left frontal lobe precentral gyrus.

	Paradigm	Cerebrum	Cluttering Group				Stuttering group			
	1/2/3	R/L	X	y	Z	Z value	X	y	z	Z value
Primary Motor Cortex	1	L	-46	-2	8	6.97	-54	-8	30	8.43
	2	L	-50	-16	40	8.69	-50	-12	38	8.74
	3	L	-48	-14	38	11.24	-50	-10	36	6.84
Premotor cortex	2	L	-58	-8	38	6.73				
	3	L	-52	-8	36	7.75				
	1	R					-58	-8	36	9.3
	3	R					60	-2	24	4.25
Thalamus, VLN	3	L	-12	-12	8	4.35				
Putamen, LN	3	R	26	-12	8	3.78				
FL Precentral G	3	L	-50	5	26	9.42				
TL Sub-G	2	L					-34	-50	22	7.47
Dorsolateral PFC	1	R					58	2	26	6.73
Subcentral area, PCG	1	R					58	-14	18	7.48

Table 3. BOLD activations (MNI coordinates) for the Active > Baseline contrast for PWC (Cluttering Group) and PWS (Stuttering group). Abbreviations: R, right; L, left; VLN, ventral lateral nucleus; LN, lateral nucleus; FL, frontal lobe; G, gyrus; TL, temporal lobe; PFC, prefrontal cortex; PCG, post central gyrus. Main effects across groups are reported at $P < 0.05$, FDR corrected. Group interactions are reported at $P < 0.001$ unless indicated otherwise. Minimum cluster size 10 voxels.

3.2.2. *The stuttering group* (see Table 3.)

Significant left cerebrum activation was located in primary motor cortex (all paradigms) and right premotor cortex (paradigm 1 and 2). Activation of the dorsolateral prefrontal cortex areas and the subcentral areas of the post central gyrus, both located in the right cerebrum, was seen in paradigm 1. Left temporal lobe subgyral activation was only seen in paradigm 2. Basal ganglia activation in stuttering was not significantly different compared to other activation levels in PWS.

3.2. Between group activation differences

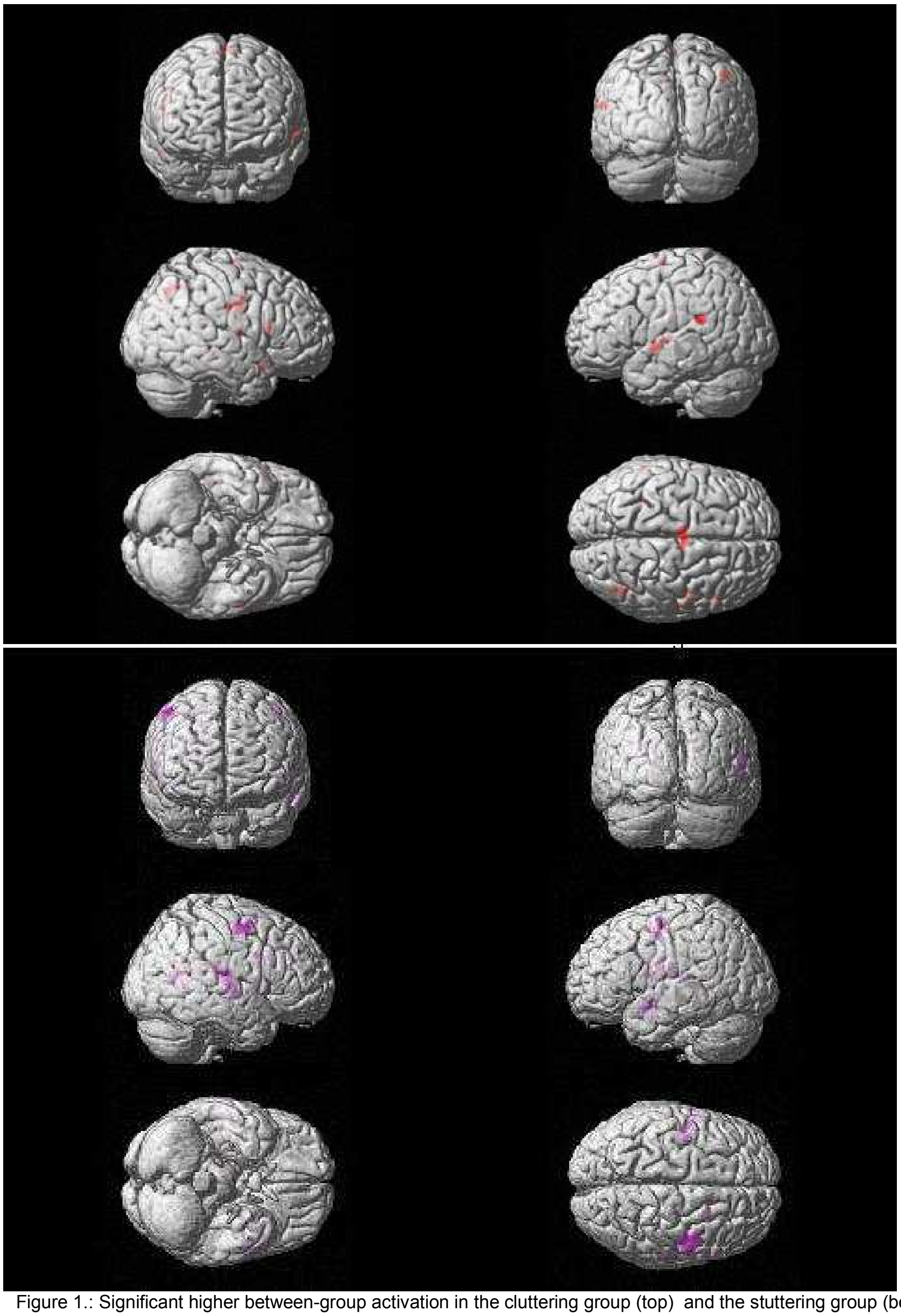


Figure 1.: Significant higher between-group activation in the cluttering group (top) and the stuttering group (bottom) in paradigm 1

3.2.1. **Increased activation: cluttering group over the stuttering group** (see Table 4.)

Paradigm 1 (see Figure 1.)

Temporal and sub lobar activation of the superior temporal gyrus of the insular cortex, the lentiform nucleus of the putamen, the ventral lateral nucleus of thalamus of the left cerebrum and right cerebrum precentral gyrus, inferior frontal gyrus, sub-lobar extra nuclear and temporopolar activation was significant higher in the AWC group compared to the AWS group.

Paradigm 2 (see Figure 2A.)

Significant higher left frontal lobe activation levels were registered in the AWC group in precentral gyrus and temporal lobe sub gyral area compared to AWS. Furthermore, significant higher activation of the inferior frontal gyrus and secondary visual cortex of the right frontal lobe was seen.

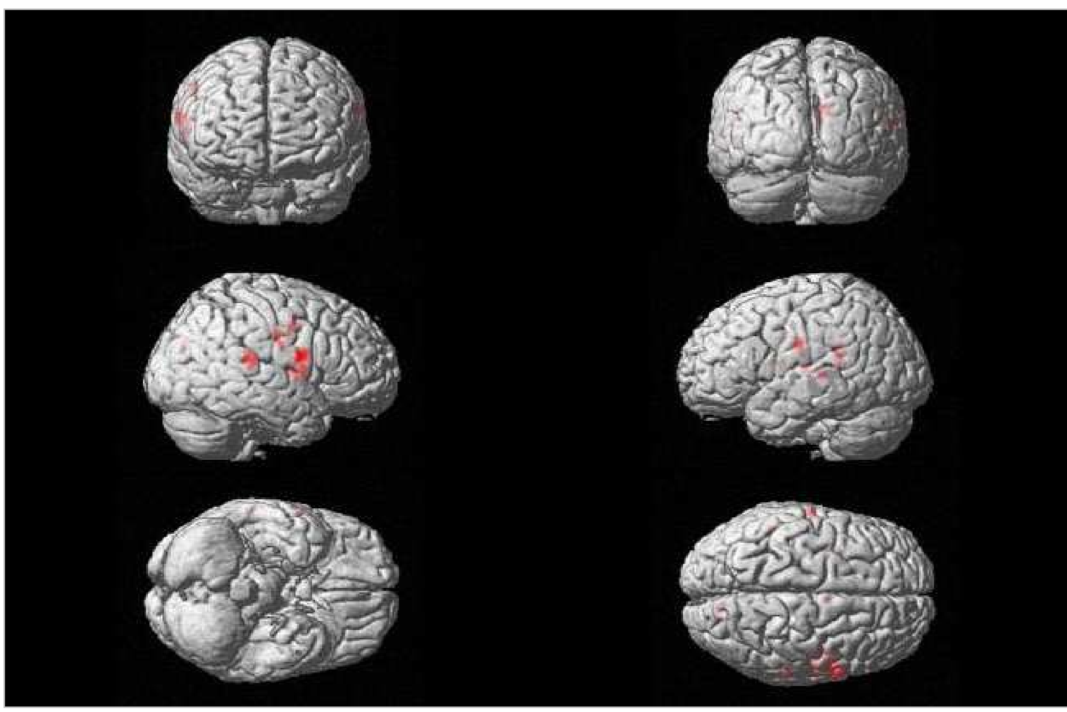


figure 2A

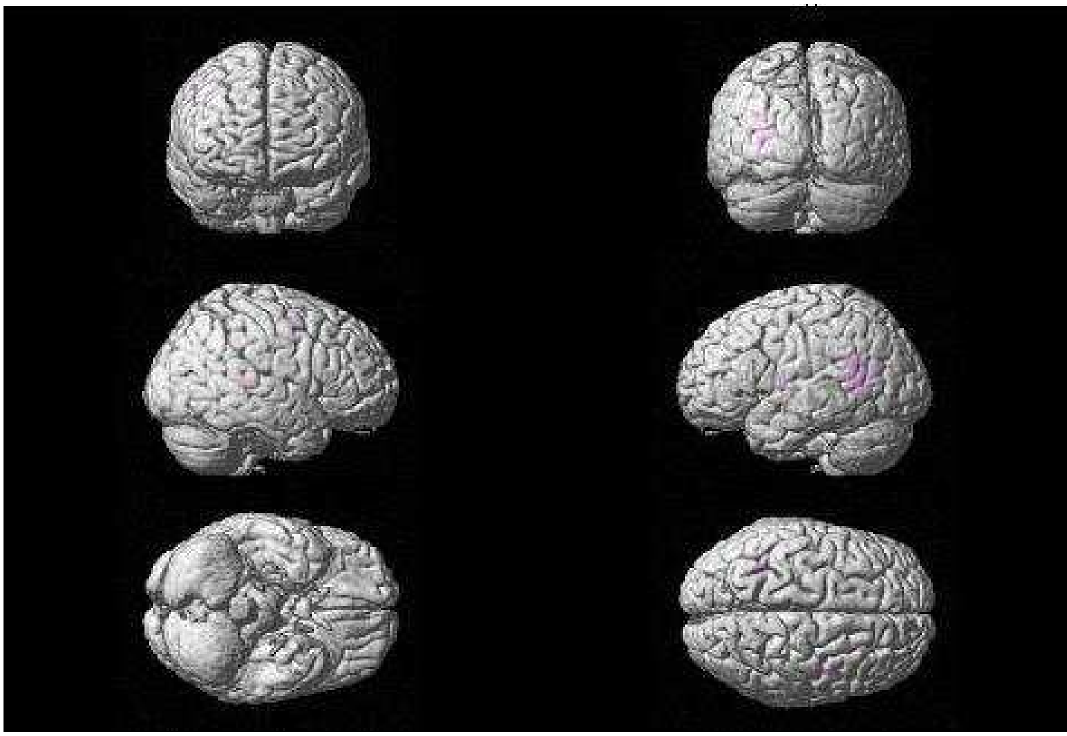


figure 2B

Figure 2.: Significant higher between-group activation in the cluttering group (figure 2A) and the stuttering group (figure 2B) in paradigm 2.

Paradigm 3 (see Figure 3.)

Significantly higher left frontal lobe activation levels were registered in the AWC group in precentral gyrus and sub gyral area compared to AWS (see Figure 3).

		Paradigm	Cerebrum	PWC > PWS				PWS > PWC			
		1/2/3	R/L	x	Y	z	Z value	X	y	Z	Z value
Frontal lobe	**PMC**	3	R					50	-12	46	3.6
	SFG	1	R					28	14	54	3.8
	Precentral G	3	L	-28	-16	40	3.9				
		1	R	44	-4	34	4.2				
		2	R	52	2	10	3.4				
		3	R	48	-16	32	5.0				
	IFG	1	R	50	20	16	3.5				
		2	R	18	32	4	3.4				
Temporal lobe	**Middle TG**	3						-58	0	-16	3.4
	Sub-G	2	L	-32	-4	-36	3.2				
		3	L	-42	50	-2	3.6				
		1	R					26	18	26	4.2
Basal ganglia	**Insular Cortex**	1	L	-56	-42	20	3.6				
	Thalamus, VLN	1	L	-12	-16	4	3.1				
	Putamen, LN	1	L	-26	-12	6	3.3				
	L. Globus pallidus	1	R					20	- 6	6	4.1
	M. Globus pallidus	3	L					-14	-6	0	4.8
Sub lobar	**Extra nuclear**	1	R	36	-4	-8	3.6				
		1	R					-2	-14	12	3.0
		1	L					-34	0	-10	3.4
Temporal lobe	**SG**	1	L					-34	-54	20	3.2
		2	L					-34	-50	24	3.5
	Temporopolar area	1	R	54	12	-20	3.3				
		2	R					44	-38	12	3.1
Occipital lobe	**Cuneus**	2	R	16	-80	26	4.5				

Table 4. BOLD activations (MNI coordinates) for the group interaction contrasts for PWC > PWS group and PWS > PWC group. Abbreviations: 1, paradigm 1; 2, paradigm 2; 3, paradigm 3; R, right; L, left; VLN, ventral lateral nucleus; LN, lateral nucleus; FL, frontal lobe; G, gyrus; TL, temporal lobe; PFC, prefrontal cortex; PCG, post central gyrus; STG, superior temporal gyrus; and SFG, superior frontal gyrus. OL, occipital lobe. Main effects across groups are reported at $P < 0.05$, FDR corrected. Group interactions are reported at $P < 0.001$ unless indicated otherwise. Minimum cluster size 10 voxels.

3.2.2. **Increased activation: stuttering group over the cluttering group** (see Table 4.)

Paradigm 1 (see Figure 1.)

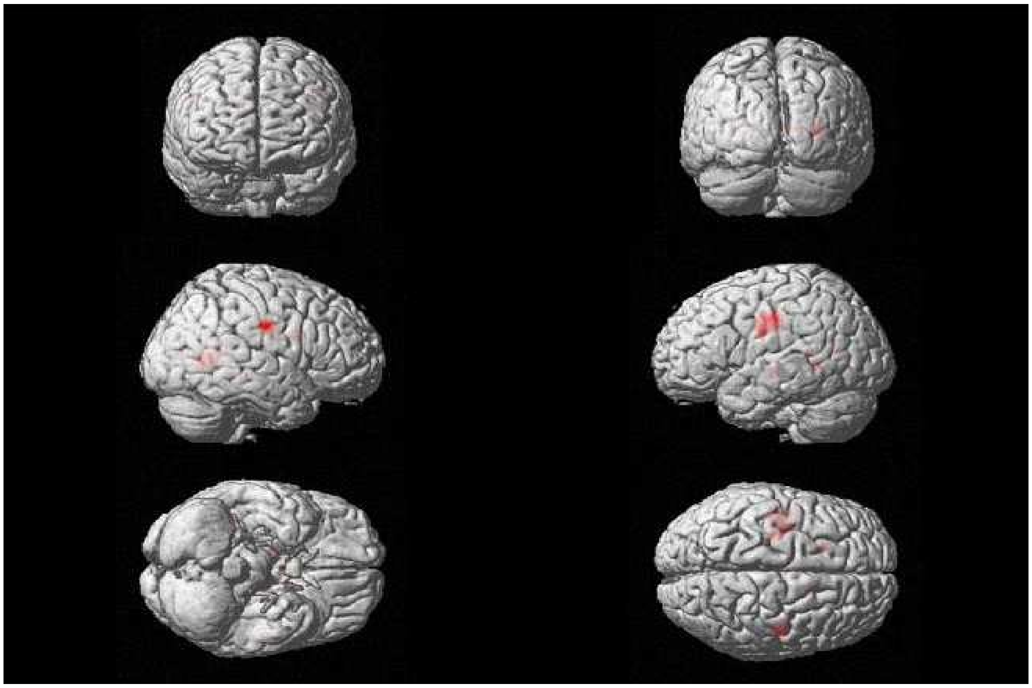
Left and right sub-gyral temporal lobe and right superior frontal gyrus activation differentiated AWS from AWC.

Paradigm 2 (see Figure 2B.)

Left superior gyrus temporal lobe activation and right temporal area activation differentiated AWS from AWC.

Paradigm 3 (see Figure 3.)

Right primary motor cortex, left sub-lobar activation in the lentiform nucleus of the medial globus pallidus and left middle temporal gyrus activation differentiated AWS from AWC.



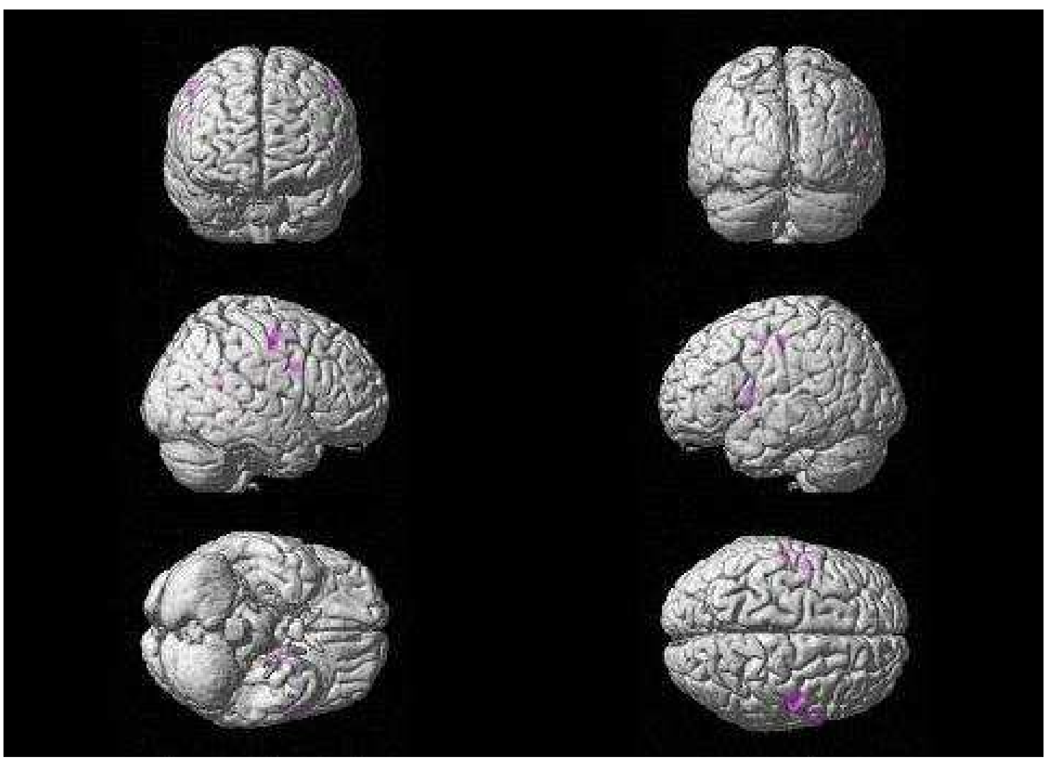


Figure 3.: Significant higher between-group activation in the cluttering group (top) and the stuttering group (bottom) in paradigm 3.

4. Discussion

The purpose of this study was to see if differences in brain activity involved in motor control and coordination during the performance of a range of spoken tasks (increasing in linguistic or motor complexity) exist in AWC compared to AWS. Specifically the activity level of the (ventral) pre-motor cortex (of the non-dominant hemisphere), basal ganglia centres and primary motor cortex was expected to be different in AWC compared to AWS. Results indicate that differences in activation levels of primary motor and pre-motor areas between adults who clutter or stutter exist. Furthermore evidence was found of different activation levels in basal ganglia centres. Although in comparing two disorder groups only relative differences between groups can be studied and therefore interpretations of these findings regarding underlying neurocognitive processes should be made with high caution, the results suggest that brain activation in cluttering is affected by rate and linguistic complexity in a different manner compared to stuttering.

The participants in this study were selected upon highly sensitive inclusion and exclusion criteria. In the cluttering group only people with a fast and/or irregular speech rate in combination with either, word or sentence structure errors, pausing errors or a high frequency of normal disfluencies were included. In the stuttering group only people who stuttered moderate to very severly and had a moderate to slow speech rate were included. People with a mixture of both cluttering and stuttering symptoms were excluded from the study as were people with language disorders, or dyslexia. Although controls are not included in this study, it is assumed that different brain activation levels in important speech motor areas of the brain of these two disorder groups can be interpreted as images of differentt underlying neurocognitive processes. Further research involving pure cluttering, pure stuttering and fluent individuals can contribute to a better understanding of the neurocognitive process in cluttering and stuttering.

In using the three different paradigms PWC showed speech related BOLD activity predominantly in the frontal lobe and sub-cortical nuclei, as is known for stuttering and other speech related brain areas. In a dynamic paradigm with both motor speech and language components paradigm 3 was expected to be complex on both linguistic and motor demands. On the assumption that stuttering is a motor disorder the activation level of primary motor cortex was expected to be higher in paradigm 3 compared to the levels of activation in paradigm 1 and 2. Results indicate that paradigm 3 showed a high level of primary motor activation in both PWC and PWS. But where activation level in cluttering gradually increased across tasks, in the stuttering group activation level remained relatively stable (see t-values in Table 3). An interpretation of these differences is rather difficult without information on activation levels in fluent controls. In a further study where controls are included these results can be studied in more detail.

The dominance of one of the cerebral hemispheres over the other hemisphere has been argued to be necessary for the coordination of motor acts used to speak fluently (Packman & Attanasio, 2004; Travis, 1978). A lack of dominance causes interruptions of speech movements, a-synchronisation leading to dysfluency (Boey, 2008). Group contrasts indicated that AWC showed higher activity in right hemisphere precentral gyrus, inferior frontal gyrus and left insula. The precentral gyrus takes part in the higher aspects of motor control and the planning and execution of behavior, tasks that require the integration of information over time. Higher activation of right precentral gyrus was reported earlier for AWS (Neuman, 2003; Viswanath, 1999; Watkins et al., 2008) and interpreted as a compensation stategy to regain fluency. The activation level of the right precentral gyrus in the AWC group was found to be even higher compared to the AWS group. The left inferior frontal gyrus looms large linguistically. This gyrus is the general location of Broca's area and is assumed to be crucially involved in phonological production. In auditory judgment of the production of the complex words outside the scanner PWS produced little to no smooth flow errors or sequencing errors, while the cluttering group produced high levels of both smooth flow and sequencing errors (see Table 1). In many models, right hemisphere activity is seen to represent compensatory behaviour (De Ridder, 2007; Preibisch, Neuman, Raab, et al., 2003; Sommer, Koch, Paulus, Weller & Buchel, 2002). The extra activation of the right homologue inferior frontal gyrus (IFG) and the precentral gyrus (PG) in the AWC group can be an indication of a compensatory activation for left IFG and PG malfunctioning. Results assume that increased activation in the cluttering group compared to the stuttering group indicates a compensatory activation of the non-dominant hemisphere in the cluttering group in an attempt to regain linguistic fluency. Further research, in which results of the two disorder groups are compared to fluent controls are needed to confirm this assumption.

In addition, significantly higher activation of the right secondary visual cortex (cuneus) was registered in the AWC group compared to the AWS group (paradigm 2). While participants looked at a black screen the AWC group possibly recalled the visual image of the word while speaking. In repeating syllable strings recall of word images is not necessary. It can be hypothesized that in PWC parts of the visual information remains active while producing the syllable strings. This phenomenon is described auditorily by van Zaalen, Wijnen and Dejonckere (2009a) for syllable production: already produced syllables remain active and where unintentionally added in later words (coarticulation) with comparable syllable structures. Alm (2007) discussed this phenomenon in relation to basal ganglia inhibitory problems. Whether this inhibition problem is indeed related to malfunctioning of basal ganglia centres needs further research.

Finally, the current research did not include a control group, with the key research questions addressing potential CNS processing differences between those who stutter and those who clutter. In following research adding a control group can serve important information regarding how the PWC differ from fluent controls in right hemisphere activation.

CONCLUSION

Differences in brain activity involved in speech motor control during the performance of a range of spoken tasks (increasing in linguistic or motor complexity) in AWC compared to AWS were present in an fMRI study. Both groups showed speech related BOLD activity predominantly in the frontal lobe and sub-cortical nuclei. Group contrasts indicated that AWC showed higher activity in right hemisphere precentral gyrus, right inferior frontal gyrus and left insula. AWS showed higher activation in right primary motor cortex, temporal lobe and globus pallidus. Findings tentatively suggest deficits in different parts of the brain are responsible for cluttering and stuttering. Further brain imaging studies in the field of cluttering should be addressed to cluttering, stuttering and fluent controls in order to interpret the underlying neurocognitive processes responsible for these findings.

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General discussion

General discussion

This study had two main objectives: (1) clinical, diagnostic classification of the syndrome of cluttering and particularly the difference in symptomatology between cluttering and stuttering and between cluttering and speaking problems associated with learning disabilities; (2) to contribute to a (neurolinguistic) model of cluttering that provides a coherent explanation for the observed symptomatology and elucidates the difference between cluttering and stuttering. For that purpose diagnostic instruments used in stuttering assessment were adapted to cluttering and newly developed assessment instruments to identify cluttering were developed and validated in a large group of disfluent speakers in the age range 6;6 – 50 years. Determining normative values for speech/language characteristics in cluttering were compared to stuttering, fluent controls and persons with speech impairment related to learning disability (LD) also provided a better understanding of some aspects underlying the different neurolinguistic processes of fluency disorders. An fMRI project confronted brain activation data in persons diagnosed with either pure cluttering (PWC) or pure stuttering (PWS). In this fMRI study the question was whether cluttering and stuttering display different neurolinguistic processes, when performing speech tasks that call upon increasing demands on speech motor and linguistic skills, was addressed.

In the first part of this discussion the results of different experiments will be considered in relation to each other. Firstly the speech and language characteristics of persons who clutter or stutter are reviewed. This description is followed by a discussion on the usefulness and validation of test instruments adjusted to cluttering. Aspects of language formulation in the cluttering group will be discussed in relation to language production of children with learning disabilities. In the second part conclusions regarding the cluttering syndrome (functional, psychological and possibly neurolinguistic) will be drawn and related to Levelt’s (1989) language production model.

Part I: Speech and language characteristics of cluttering and stuttering.

St. Louis, Raphael, Myers and Bakker (2003) defined cluttering as:

> *“a syndrome characterized by a speech delivery rate which is either abnormally fast, irregular, or both. In cluttered speech, the person’s speech is affected by one or more of the following: (1) failure to maintain normally expected sound, syllable, phrase, and pausing patterns; (2) evidence of greater than expected incidents of disfluency, the majority of which are unlike those typical of people who stutter”.*
> (St. Louis, Raphael, Myers & Bakker, 2003).

In order to identify cluttering, an answer is needed to the question: 'What is an abnormally fast or irregular speech delivery rate?' To our knowledge, there was so far no clinical or scientific consensus on this issue. Consensus regarding "*maintenance of normally expected sound, syllable, phrase timing, and pausing patterns",* as well as the number of "*expected incidents of disfluency*" was lacking as well. In order to establish norms on these speech language characteristics data was gathered on: articulatory rate; articulatory accuracy and smooth-flow, frequency and type of normal disfluencies. Conclusions on different speech variables in PWC and PWS are displayed in Table 1.

In collecting the data the test sequence for all participants was: (1) monologue; (2) reading; (3) retelling wallet-story; (4) speech motor control on syllable (OMAS, Riley, 1984) and word level (SPA, Screening Pittige Articulatie, Van Zaalen et al., 2009a). In the next section data on articulatory rate, word structure and incidents of disfluencies in PWS and PWC will be discussed, followed by a presentation and discussion of the test instruments used.

	PWC		**PWS**	
	M	*SD*	*M*	*SD*
MAR monologue	5.3	0.7	3.7	0.8
MAR reading	4.5	1.7	4.0	1.0
MAR retelling	4.9	0.9	3.7	1.5*
MAR OMAS	6.0	1.2	4.2	1.2*
MAR SPA	4.3	0.8	4.7	1.2
Accuracy SPA	1.5	1.0	0.8	0.9*
Coarticulation SPA	2.3	1.6	6.9	5.6**
Flow SPA	3.1	1.9	1.3	1.2*
Sequencing SPA	4.1	3.5	1.1	1.1*
Rate SPA	7.8	2.9	7.6	2.0
RD Monologue	6.4	3.9	0.4	0.5**
RD Retelling	7.6	4.4	2.0	2.0**
RD Reading	2.1	2.7	0.4	0.4
SSI-3 scores	No stuttering- light stuttering		No stuttering – very severe stuttering	

** significant difference p< .001
* significant difference at p< .05

Table 1.: Speech characteristics in persons who clutter (PWC) and persons who stutter (PWS), means and standard deviations. Abbr.: MAR: mean articulatory rate; SPA: Screening Pittige Articulatie (Van Zaalen et al., 2009a); RD: ratio disfluencies; SSI-3: Stuttering Severity Instrument-3, Riley (1994); PCI-revised: Predictive Cluttering Inventory revised (Van Zaalen et al., 2009c).

Articulatory rate

Articulatory rate, in stuttering research, is usually calculated in syllables per second or phonemes per second by analysing only perceptually fluent utterances. Articulatory rate measures are intended to reflect how fast sound segments are produced in stretches of speech that have no pauses (Hall, Amir, & Yairi, 1999; Pindzola, Jenkins, & Lokken, 1989). Perceptually fluent utterances are defined as those utterances that do not contain "within- or between-word disfluencies, hesitations, or pauses greater than 250ms" (Yaruss, Logan & Conture, 1994, p. 221). Mean Articulatory Rate (MAR) was defined as the mean of five rate measures in minimally 10 to maximally 20 consecutive syllables in perceptually fluent speech without pauses.

When PWC can be differentiated from PWS by a fast or irregular speech delivery rate, it is assumed that the MAR of PWC is significantly higher or more irregular compared to PWS. Although PWC had a higher MAR compared to PWS in all speech tasks (except for the SPA-test) the differences were only significant in retelling a memorized story and OMAS (see Table 1.).

Based on our empirical data, a fast speech delivery rate, expressed in MAR, is not sufficient as a differential diagnostic criterion in all speaking tasks (see Table 1.). It is possible that speech delivery rate is perceived as fast, while values within normal limits are found when measured (St.Louis et al., 2007). At least three explanations can be given for the phenomenon that speech delivery rate is perceived as fast, while articulatory rate is within normal limits.

First, speech delivery rate can be influenced by extra or extended pauses. Pausing 'errors' within and between sentences occur in PWC when pausing is not done according to linguistic rules (too short/long, too less/often) (Ward, 2006). PWS can experience extended pauses in response to a block. The number or duration of pauses is not taken into account in my work.

Secondly, PWC sometimes produce unintelligible spurts of speech in which it is difficult to objectively determine how many and which syllables and phonemes have been realized. To avoid overly subjective assessment, the articulatory rate was calculated on the basis of the number of syllables that should have been realized [for the citations forms of words]. Spurts in speaking rate are often unintelligible and therefore not included in the MAR. In doing this a major characteristic of cluttering is left out from the articulatory rate measurements.

Finally, a speech delivery rate can be perceived as fast due to disfluencies. A high amount of normal disfluencies in combination with a fast speech rate can have a bad influence to speech intelligibility (Sick, 2004; Van Zaalen & Winkelman, 2009).

In future research on cluttering it is recommended to develop an analysis technique in which unintelligible spurts and pauses are included in rate measurements. One way to do this is by calculating the percentage intelligible and unintelligible speech time. Another way of doing this is by measuring the duration and accuracy (pausing at linguistically appropriate points) of pausing in different speech tasks. It is assumed that in combining rate, pausing and intelligibility measurements more information will be gathered on the effectiveness of speech delivery in PWC.

Word structure errors

In order to classify errors in word structure, the SPA (Screening Pittige Articulatie) was developed. In an elicitation procedure, three repetitions of ten multi-syllabic words at a fast speech rate were obtained. In the SPA, scores were obtained on five different categories: (a) Accuracy, (b) Coarticulation, (c) Flow, (d) Sequencing, and (e) Rate. Problems in sound accuracy (distortions or substitutions of voicing and devoicing) were considered to be an indication of problems in adjusting voicing to articulatory movement: an indicator of difficulty realizing adequate voice-onset time. Coarticulation is the gradual transition from one speech movement to the next. In the SPA-test telescoping syllables, and within sequence pausing were counted as coarticulation errors. Flow is the gradual stressing and rhythm of the sequence. Errors in flow, for example, may include changes in the stress pattern for the sequence. Sequencing errors were scored when a person made sound order errors within syllables.

PWC produced significantly more errors of distortion and voicing, flow and sequencing compared to PWS. These results are in line with the hypothesis that PWC can be differentiated from PWS by the number of word structure errors in fast speaking rate. Analysis of word structure errors in other speaking rate conditions is needed to answer the question if these errors are related to the speaking rate or express basic problems in phonological encoding in PWC. In future research it is advised to study word structure errors in extended recordings of spontaneous speech (3-5 minutes) and reading aloud. Rate scores on the SPA did not differentiate between PWC and PWS. This can be explained by the extra and extended pausing in the PWS group and repetition or addition of syllables in the PWC group. Persons who clutter are able to maintain a slow(er) speech rate for a short period of time. It is expected that recordings of 3-5 minutes will, comparatively speaking, exhibit a higher percentage word structure errors compared to recordings of 1-2 minutes.

Disfluency ratio (RD)

According to the 1998 ASHA guidelines on fluency: "*the generic term disfluency refers to breaks that are normal, stutter-like, or ambiguous"* (ASHA, 1998). *"The most commonly regarded normal disfluencies are: hesitations or long pauses for language formulation; word fillers or filled pauses or interjections; and phrase repetitions" (ASHA, 1998).* Stutter-like disfluencies refer to the private, personal experience of an involuntary loss of control by the person who stutters (Shapiro, 1999). *"The most commonly regarded stuttering-like disfluencies are: part-word repetitions; prolongations; blocks (silent fixations/prolongations of articulatory postures) or noticeable and unusually long (tense/silent) pauses at unusual locations to postpone or avoid; and any of the above categories when accompanied by decidedly greater than average duration, effort, tension, or struggle (ASHA, 1998)".*

According to the St. Louis et al., (2003) definition of cluttering, an evidence of greater than expected incidents of disfluency (the majority of which are non stutter-like disfluencies) was to be expected in PWC. A norm for '*number of expected incidents of disfluency*' in different speaking situations is missing. A way of analysing the disfluencies is looking at the ratio of normal disfluencies/stutter-like disfluencies (=disfluency ratio). Data indicated that the disfluency ratio differentiated between PWC and PWS in monologue and retelling a story. PWC had a higher incidence of normal disfluencies compared to stutter-like disfluencies. In reading aloud the disfluency ratio was not significantly different between PWC and PWS. Both PWS and PWC produced a low number of disfluencies in reading aloud. A relatively low incidence of stutter-like disfluencies was expected in the cluttering group. PWC produced at least 2.5 times more normal disfluencies compared to stutter-like disfluencies (see Table 1). Data on incidents of disfluencies in other diagnostic groups were needed to conclude if the number of expected incidents of disfluencies were indeed higher in PWC.

Data on retelling a memorized story showed a number of normal disfluencies and a ratio disfluencies in LD-children and controls comparable to what was found in CWC (see Table 2.). Results on CWS replicated the observation done in the population of PWS (all ages), (Van Zaalen, Wijnen & Dejonckere, 2009c).

	CWC	CWS	LD	Controls
Retelling				
- RD	8.4 (*SD*=4.6)	2.2 (*SD*=2.5) *	12.0 (*SD*=9.0)	8.1 (*SD*=4.8)
- % NDF	10.4 (*SD*=7.8)	5.0 (*SD*=3.1) *	14.7 (*SD*=14.1)	10.0 (*SD*=6.3)

* = significant difference at $p < .05$

Table 2.: Ratio disfluencies (RD) and percentage normal disfluencies (NDF) in retelling a memorized story in children who clutter (CWC), have learning disabilities (LD) and controls (Van Zaalen, Wijnen and Dejonckere, 2009b).

We can therefore conclude that this study supports the hypothesis that PWC and PWS can be differentiated by the number of normal disfluencies. No supportive evidence is found for this hypothesis in relation to either fluent controls or LD-children.

Language production disturbances in cluttering

Since Weiss described cluttering as a result of a central language imbalance, various authors discussed the contribution of language problems in fluency disorders (Damsté, 1984; Freund, 1952; Luchsinger, 1963; St. Louis, 1992; St. Louis et al., 2003, 2007; Scripture, 1912; Van Zaalen, Wijnen & Dejonckere, 2009a; Voelker, 1935; Ward, 2004, 2006; Weiss, 1964, 1968). Freund (1952) and Luchsinger (1963) described cluttering as a dysphasia-like disorder. In contrary, DeHirsch (1961) described cluttering as a disruption of motor integration, resembling dyspraxia. The linguistic problems of persons who clutter were taken into account by van Riper (1982) when he described his track II stuttering (stuttering with a severe cluttering component). In 1984 and 1988, Damsté described three different types of cluttering: dysrythmic, dysarthric and dysphasic cluttering. According to Damsté, dysphasic cluttering was characterized by sentence structure errors and sentence revisions. Language problems in cluttering were furthermore confirmed by St. Louis (1992, p. 49) when he defined cluttering as ‘a speech-language disorder’ and by Daly, who defined cluttering as “*a speech and language processing disorder resulting in a fast, dysrythmic sometimes disorganised and often unintelligible speech, where accelerated speech is not always present but...* problems*... in language formulation almost always..* are*”* (Daly, 1992, p.107). Data of retelling a memorized story experiment (see Chapter 5) with children who clutter (CWC) or have a speech impairment related to learning disability (LD-children) indicates differences in the underlying processes of language disturbances between children with cluttered speech and those with learning disabilities. The findings were interpreted in terms of Levelt’s (1993) language production model. CWC produced as many primary and secondary plot elements (on average) as controls did. This indicates that CWC are able to repeat a given story structure and does not support problems in the conceptualiser in cluttering. CWC produced correct sentence structures as controls did, with this remark that CWC used ‘editing words’ (interjections, revisions, word and/or phrase repetitions) in order to overcome formulation problems. This finding gave some indication that language planning disturbances in CWC arise due to insufficient completion of the editing phase of sentence structuring (the Formulator).

Language production of LD-children was disturbed by problems at the conceptualiser and formulator stages. LD-children produced significantly more errors in story structure, less primary and secondary story components and more sentence structure errors (at first and second attempt). These observations are according to data provided by Wigg and Semel (1984). The role of language production in cluttering in further discussed in part II of the discussion.

Assessment and screening instruments developed for or adapted to cluttering

In this study an instrument used in stuttering assessment, the Stuttering Severity Instrument-3 (Riley, 1984), is adapted to cluttering, by including normal disfluencies and the ratio disfluencies. In the Stuttering Severity Instrument-3 normal disfluencies are not counted when speech production is observed. In counting the normal disfluencies and calculating the disfluency ratio (normal disfluencies/stutter-like disfluencies) a differentiating characteristic between cluttering and stuttering was established. However, the ratio disfluencies in persons who clutter appeared to be not significantly different from fluent speakers or LD-children. Cluttering has to be regarded as a syndrome with more than one characteristic.

A screening instrument for cluttering behaviour, the Predictive Cluttering Inventory (PCI), was validated. The original PCI was neither sensitive nor specific enough to detect cluttering. Van Borsel et al., (2008) drew the same conclusion in a group of participants with Down-syndrome. The sensitivity level of the revised PCI is sufficient. Item clarification and examiners instructions can raise the predictive value of the screening instrument.

An assessment instrument, ‘the wallet story’, was developed to analyse language production at the levels of the conceptualiser and the formulator. Although this instrument is not restricted to cluttering it provides useful information on language production in a speech task different from reading aloud and spontaneous speech.

Concluding

Data gathered in this study support the hypothesis that objective measures of articulatory rate, type and frequency of disfluencies and errors in word or sentence structure differ between PWC and PWS. Support is found for the assumption that in cluttering, high or highly variable rate engenders difficulties in language formulation and that these disturbances diminish when speaking rate is reduced or linguistic demands (complexity) decrease(s). Syntax and story structure errors in LD-children are assumed to reflect a lower level of language development.

Part II: underlying neurolinguistic processes.

In the first part of the general discussion differential diagnostics between cluttering, stuttering and speech impairment related to learning disability are discussed. In this part of the discussion underlying neurolinguistic processes in cluttering, stuttering (and speech impairment related to learning disability) will be considered in relation to brain activation data and the language production model of Levelt (1989).

Cluttering and stuttering: two different disorders.

In an fMRI experiment the question of possible differences in brain activity involved in motor control and coordination during the performance of a range of spoken tasks (increasing in linguistic and motor complexity) in PWC compared to PWS was addressed. FMRI data was collected in adults who stutter but do not clutter (AWS) and adults who clutter but do not stutter (AWC). The tasks involved pronouncing words of increasing complexity. A block design with a 40 second on-off period that alternated viewing symbols with reading aloud words was used.

In the brain imaging study with participants diagnosed with ‘pure’ stuttering and ‘pure’ cluttering both PWC and PWS showed speechrelated BOLD activity predominantly in the frontal lobe and sub-cortical nuclei. Although the activation was registered in the same areas, there were differences in levels of activation between PWC and PWS that possibly refer to different neurocognitive processes in these disorders.

Significantly higher frontal lobe activation levels were registered in the PWC group compared to PWS in precentral gyrus, inferior frontal gyrus (IFG) and temporal lobe sub gyral area. The precentral gyrus takes part in the higher aspects of motor control and the planning and execution of behavior, tasks that require the integration of information over time. Higher activation of right precentral gyrus was reported earlier for PWS and interpreted as a compensation stategy to regain fluency (De Ridder et al., 2007; Neumann et al., 2003; Preibisch et al., 2003; Sommer et al., 2002; Viswanath, 1999; Watkins et al., 2008). The activation level of the right precentral gyrus in the PWC group was found to be even higher compared to the PWS group. It is unknown how the activation level relates to that of fluent controls.

The IFG is the general location of Broca's area and is assumed to be crucially involved in phonological and syntactical processing. Or as Menenti et al., (2007) described it "*for unifying information from different resources into coherent representation of a multi-word utterance*". Results suggest that the increased right hemisphere precentral and IFG activation in PWC, compared to PWS, represents compensatory behaviour in an attempt to gain (linguistic) fluency. It is tentatively hypothesised that the overactivation in PWC can be a sign of defective automated language production processes in PWC and that this extra activation is needed to complete language coding processes. Further research, in which results of the two disorder groups are compared to fluent controls are needed to confirm this assumption.

The findings also tentatively suggest that different neural mechanisms are responsible for cluttering and stuttering and that cluttering and stuttering are two independent disorders of fluency with different pathogeneses. Therefore, it is reasonable to hypothesise that PWS or PWC display different neurocognitive processes when performing speech tasks.

Chang et al., (2009) reported greater activation in the right STG, insula, putamen, and precentral motor regions in PWS during speech and non-speech production compared to controls. In our experiment PWC exhibited greater activation compared to PWS in these areas. In comparing two disordered groups only relative differences between groups can be studied. It is possible that the observed higher activation in the PWC group was in correspondence with expected activation in fluent speakers. Some indications for that assumption are found in the study of Davidow et al., (2009). Comparison of results in our study to other studies including PWS is very difficult because the speech task used in this experiment was unique. Therefore, follow-up studies on cluttering should include fluent subjects performing the same tasks.

Cluttering and language production

The next part of this general discussion focuses on a tentative theory, based on insights in the structure of the language production system, which can explain the main characteristics– both core symptoms and secondary phenomena - of cluttering. Core symptoms are those characteristic features a person who clutter is recognized by: *"a too high or too irregular speech delivery rate, failures to maintain expected sound, syllable, phrase, and pausing patterns, often manifested by articulatory errors, and/or a higher than expected incidence of normal disfluencies*" (St. Louis et al., 2003). Secondary phenomena refer to global phenomena, characteristic for the cluttering syndrome. They can occur in daily life, but also during exceptional circumstances (e.g., experiments). The first secondary phenomenon of cluttering is variability depending on social context. It is reported by different authors that even very severe PWC show moments of clutter free speech (Daly, 1986; Ward, 2006; Weiss, 1964). Reduction and increase of disfluencies influenced by the social context (communication partner), content of the message, emotional condition, etc. are mentioned in clinical handbooks (e.g. Myers & St.Louis, 1992; Van Zaalen & Winkelman, 2009; Ward, 2006) as characteristic for cluttering.

A second secondary phenomenon is the temporary reduction of cluttering by, on the one hand, changes in speech behaviour (e.g. lowering speech rate, increased attention to articulatory movements), and on the other hand manipulation of auditory feedback (St.Louis, 1992; Ward, 2006). For instance, language production disturbances in CWC can be similar to developmental language disorders (e.g. specific language impairment (SLI)). But, in contrast to children with SLI, language production disorders (i.e. errors in grammatical or phonological encoding) in CWC decrease when speaking rate is decreased.

A third secondary phenomenon is the lack of awareness of cluttering symptoms. PWC are usually aware that their speech is less intelligible or fast, but are not aware of the cluttering moments during speech production. This phenomenon is described by many authors based on clinical observation (St. Louis et al., 2007; Van Zaalen & WInkelman, 2009; Ward, 2006; Weiss, 1964) and by some on experimental results (Van Zaalen et al., 2009d; Williams & Weener, 1996).

In speaking situations with heightened attention to speech production and in situations with a mean speaking rate below normal levels language production quality of PWC is comparable to that of fluent controls (Van Zaalen et al., 2009b). Language production in spontaneous speech of PWC can be characterized by a higher than normal level of word or sentence structure errors (St. Louis et al., 2007). When language production is fully automated it is assumed to be rarely affected by rate or linguistic complexity. It is hypothesized that language production in cluttering is insufficiently automated. In the next section the hypothesis of a defective language automatisation in cluttering is explored.

The language production system

Before language production in cluttering will be discussed, language production in general will be described, using Levelt's model of language production (Levelt, 1989). Disfluencies will be discussed within the Covert Repair Hypothesis (Kolk & Postma, 1993) and the EXPLAN model (Howel & Au Yeung, 2005).

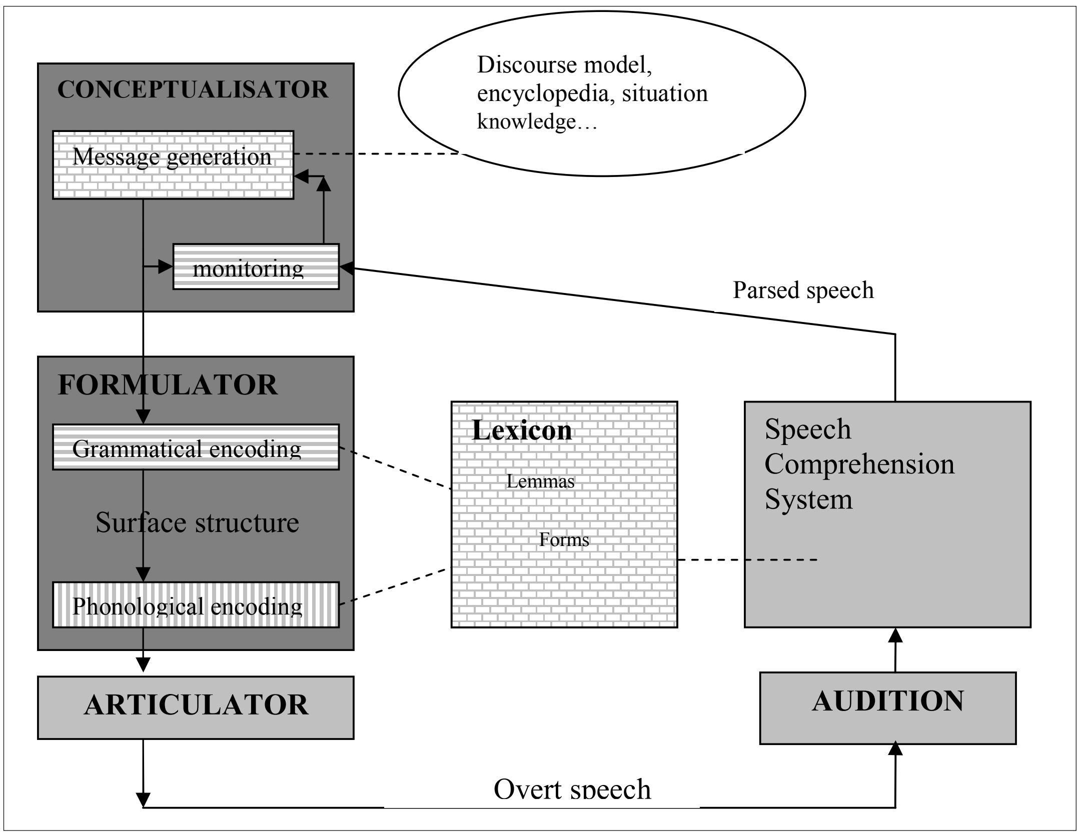

Figure 1.: Language production model (after Levelt, 1993)

In Levelt's (1989) model (as well as various others), utterance planning takes place in three steps, each of which are subserved by distinct (cognitive) modules: conceptualiser, formulator, and articulator. The first component is the 'conceptualiser', which selects and organizes information to be communicated, generally identified as the "preverbal message". The preverbal message activates the formulator. The Formulator comprises two processing devices: the grammatical encoder and the phonological encoder. The mental lexicon plays a central role in the formulator. Lexical items (lemmas) are activated on the basis of their semantic features. Once they are active, their syntactic and morphological properties are made available and the formulator encodes them into sentences. Once the lemmas are put in grammatical order (grammatical encoding), their corresponding phonological forms (lexemes) are retrieved and encoded (phonological encoding). Lexemes comprise prosodic and segmental (phonemic) information – i.e., the number and nature of the speech sounds in a word, and the position of primary (and secondary) stress. The output of the formulator is a phonological representation of the intended utterance. This phonological representation is transferred to the articulator, which transforms it into a speech motor plan.

Speakers are assumed to continuously evaluate the linguistic correctness and pragmatic adequacy of their utterances (monitoring). Levelt (1989) described three ways of monitoring: (a) within the conceptualiser: control and correction of preverbal, propositional code; (b) inner loop: internal monitoring, control and correction of the output of the formulator before it is articulated and made audible; and (c) the outer loop: perception of realized speech, and comparing it to the original intention. Note that the second and third monitoring channels rely on the speech comprehension system, i.e., the same system that is engaged when we listen to and understand *other* speakers (Postma, 2000).

The defective language automatisation hypothesis

In the above paragraphs the three processing steps (conceptualising, formulation and articulation) in Levelt's model of language production were described. Before the problems reported for PWC in relation to these steps in language production are discussed, assumptions related to cluttering, disfluencies and automatisation of language formulation will be spelled out.

Cluttering can be characterised by an exceptionally high incidence of normal disfluencies, i.e., *hesitations or long pauses, word fillers or filled pauses or interjections; and phrase repetitions" (ASHA, 1998).* On the assumption that syntactically and phonologically correct sentences can be produced in fast speech rate when the process of language formulation is automatised, I hypothesize that language formulation processes in cluttering are insufficiently automatised. When language formulation is insufficiently automatised, errors in language production can be expected. If, due to a lack of automatisation, much attention is needed to complete grammatical or phonological encoding, fewer resources remain for other processes, particularly monitoring and articulation. Consequently, a high level of language production difficulties remains.

An influential hypothesis in which disfluencies are related to processes in the language production system is Kolk and Postma's Covert-Repair-Hypothesis (Postma & Kolk, 1993). Although the Covert Repair Hypothesis (CRH) presented as a model explaining stuttering, has been argued by different authors (Howell, 2004; Yaruss & Conture 1996; Wijnen, 2000), it is possible that it is valuable in explaining cluttering. The core idea of the CRH is that all disfluencies reflect self-corrections (a.k.a. repairs). In some self-corrections the error (or 'reparandum') is overt, and it is clear how the ensuing correction changes this error into a correct string. An example is: "*You made so much noise you worke Cor? – wore? – w?- woke Corky up" (Postma, 1991)*. Following a suggestion made by Levelt, Postma and Kolk (1993) assume that disfluencies in which the utterance form is not overtly changed ("this is .. a horizontal line") reflect a covert repair (the error itself is not perceptible for the outsider). The error is detected and corrected before it is articulated. The interruption of ongoing speech and the restart are audible.

If word and phrase repetitions, revisions and hesitations, characteristic for cluttering, are repairs of detected errors, the question is, why do such errors occur so regularly in a clutterer's speech? And the second question is why do PWC not detect and repair all errors? And finally, the third question is, why do the disfluencies occur more frequently when speech rate is high? In samples of PWC limited indications were found of PWC detecting and repairing errors in running speech.

In their Explan-model, Howell and Au Yeung (2005) claim that "*fluency failures should not be seen as repairs but as a representation of a point where planning cannot keep up with output speed*". When cluttering is an expression of defective language automatisation processes, the disfluencies can be an expression of a speech plan that is not ready in time. A word-plan is normally delivered ahead of motor execution (Howell & Au Yeung, 2005). "*Planning can be put under time pressure when a segment that is difficult, and therefore time-consuming to generate, has to be prepared. This plan is required quickly as when the planned segment follow a word that is executed rapidly .... the process can fail to deliver the complete plan in time...*" (Howell, 2004, p81). When the plan is not complete, the message can not progress fluently. In this situation the speaker can retrieve the plan of a word or phrase that is recently used and execute it again (word or phrase repetition). As planning takes place in parallel with execution, the planning of the next part can continue unaffected during the repetition (Howell & AuYeung, 2005). According to the Explan-model speech motor execution can proceed when the next plan is complete. In doing this the speaker gains time to complete the speech plan.

Observations that seem to support a time gaining effect on grammatical encoding in cluttering are described by Van Zaalen et al., (2009b) when they studied syntactical correctness in sentence structures. PWC were able to produce a percentage correct sentence structures comparable to fluent controls when a sentence was judged after removal of the word- and phrase repetitions and revisions. Repairs of grammatical encoding problems are most likely to be heard as revisions (changes are made in the prior plan). Both revisions and word-phrase repetitions are observed more frequently compared to controls in PWC (Van Zaalen, 2009b,c). Based on these results it is assumed that language production in PWC is not fully automatised. When language production is not fully automated language output will be characterised by many errors and disfluencies.

Problems in phonological encoding are most likely to be heard as speech errors, and covert repairs as part-word repetitions. Some evidence of undetected errors and a time gaining effect of repetitions, indicating an automatisation problem, was found on the level of phonological encoding when syllable sequencing errors and phoneme perseverations (already produced phonemes somehow reoccur in the speech plan) were frequently observed during the SPA-test (Van Zaalen et al., 2009a).

Monitoring

Wijnen (2000), claims that PWS invest a large share of processing resources in monitoring. This, in itself, however, does not account for the primary symptoms of stuttering, notably sub-syllabic repetitions and blocks. Hence, Wijnen (2000) additionally assumes that stuttering persons focus their monitoring device on prolongations and interruptions of articulation. In doing so, they maintain an unadaptively high threshold for acceptability, as a consequence of which not only (imminent) hesitations, but even linguistically or articulatory determined temporal fluctuations are evaluated as output errors. "*This excessive and lopsidedly focussed monitoring is detrimental, rather than beneficial to speech fluency, as it engenders many unnecessary interruptions and restarts of the ongoing speech*" (Wijnen, 2000).

PWC do not always repair grammatical or phonological encoding errors in running speech, which result in word/sentence errors and errors in pauses. Lack of response to speech errors or disfluencies can be a sign of weak monitoring skills. PWC are able to monitor their speech, when attentional resources are not so much needed for encoding, for instance in producing nonsense syllables. PWC proved to be able to monitor their articulatory rate for a short period of time (Hartinger & Mooshammer, 2008; van Zaalen, Wijnen and Dejonckere, 2009c). This monitoring was evident in a consistent articulatory rate between syllables when producing strings of non sense syllables.

Myers (1992) also explained the remaining errors or disturbances in language production of PWC to limited skills in self-monitoring. In problems of self-monitoring I refer to two parameters: audition and effort. Audition refers to the ability to identify, and comprehend what has been heard. Effort refers to the amount of attention invested in monitoring.

Although audition is undisturbed in PWC, self-monitoring in speaking can nonetheless be disturbed. Several empirical observations support this. (1) PWC are unaware of their speech errors while speaking (Weiss, 1964; Myers & St. Louis, 1992; St. Louis et al., 2007; Ward, 2006), while clinical observation indicates that PWC are capable of detecting errors when listening to their own recorded speech, (Van Zaalen & Winkelman, 2009; Ward, 2006). (2) Increasing intensity of auditory feedback during speech production generally ameliorates clutterers' fluency (St. Louis, Myers & Cassidy, 1996). (3) Delayed Auditory Feedback (DAF) and Altered Auditory Feedback (AAF) temporarily improve speech quality/fluency in PWC (St. Louis, Myers, Cassidy et al., 1996). This improvement can be interpreted as a result of (temporarily) improved monitoring through DAF/AAF, because attentional resources are supported by an extra external feedback resource.

Furthermore, it is a well known fact that speech production of PWC is much better when greater effort is put into maintaining clarity (Weiss, 1964; Daly, 1986; St, Louis, 2007; Van Zaalen & Winkelman, 2009; Ward, 2006). Increased effort leads to a lower rate. When speech rate of persons without a history of speech and language disorders is lowered, disfluency is reduced and the intelligibility increases (Postma, 1991).

The effect of speech rate on the number of disfluencies in PWC has not been studied directly. Based on reported decrease in error rate (Van Zaalen et al., 2009d) it is assumed that increased effort in PWC leads to a reduction of speech rate, and increases the time to complete the encoding process. Slowing down provides more time for the selection of phonological segments, thereby reducing the possibility of an error (Packman & Attanasio, 2004). Not all PWC exhibit increased disfluency as a function of increased articulatory rate (St. Louis et al., 2003; VanZaalen et al., 2009c). This suggests that increased articulatory rate does not determine disfluency frequency with the same consistency as does decreased articulatory rate. It is assumed that lowering speech rate can result in more attention to speech and language production.

In line with the disfluencies observed in cluttering the high frequency of undetected errors can be an effect of invalid monitoring due to a lack of automatisation of language production as well. This hypothesis was supported by evidence found in an experiment with the SPA-test by Van Zaalen et al., (2009c). PWC, PWS and controls had to repeat low frequency multisyllabic words at a fast speech rate. In this experiment undetected errors in PWC (accuracy, flow and sequencing) increased significantly, compared to PWS and controls for fast speech rate and linguistic complexity.

Concluding

PWC seem to monitor insufficiently because attentional resources are used for grammatical and phonological encoding. Thus, it is hypothesized that a 'double deficit' exists in cluttering: a deficit in encoding (grammatical and phonological) due to less automatisation and as a result of that, weak monitoring, because processing capacity is used for the encoding processes.

In the above sections the role of automatisation in language formulation is described in relation to cluttering and disfluencies. In the next section the reported problems in the three steps of language formulation (conceptualiser, formulator and articulator) are described and related to the defective language automatisation hypothesis of cluttering.

Conceptualiser and cluttering

Problems in conceptualising (linearization, taking perspective and referral) in cluttering were mentioned by different authors. Mensink (1990), Myers (1992), St. Louis (1992), Teigland (1994) and Ward (2006) described that persons who clutter manifest pragmatic errors (taking perspective and referral) and communication errors (inefficient turn taking) more frequently compared to normal speakers. However, it is important to state that although these pragmatic errors are described by different authors, pragmatic skills have not been investigated directly in PWC in running speech. An experiment by Van Zaalen, Wijnen and Dejonckere (2009b) in which people had to retell a memorized story did not reveal a higher level of pragmatic errors in PWC compared to controls. In this experiment no evidence was found to support Myers (1992) and St. Louis & Myers (1995) who reported (based on clinical observations) that PWC often have difficulty producing clear and cohesive narratives. Assuming that Myers and St. Louis based their remarks on the observation of spontaneous language the difference in story structure abilities between spontaneous speech and retelling a story can be an indication of automatisation problems. An experiment in which the amount of story structure errors in spontaneous speech and retelling a story of PWC are related to each other can provide some information on the automatisation in the conceptualiser.

Formulator and cluttering

Language production difficulties in cluttering are mentioned by different authors (Becker & Grundman, 1970; Bradford, 1970; Daly, 1986; Daly, 1993; DeHirsch, 1970) and at the same time denied by several others (deFusco & Menken, 1979). Four explanations for this contradiction will be discussed. Firstly, the criteria for language problems differ across authors. Some authors think of language formulation problems to be correlated with cluttering only when they occur at all times, while other researchers mention that these language formulation problems can be present but mostly disappear at a low speaking rate. Secondly, this difference is probably for the most part created by a fundamental difference between PWC: some PWC have problems with intelligibility but are capable to produce correct sentence structures, whereas others experience problems in maintaining fluency in running speech at a fast speaking rate. Thirdly, Levelt's concept of formulation differs from the concept of formulation in researchers on cluttering. Finally, problems in phonological encoding are frequently misinterpreted as motor problems.

Problems of lexical retrieval and grammatical encoding are not seen in every PWC. However, it is hypothesized that some PWC use sentence revisions, interjections and phrase repetitions in order to gain time for grammatical encoding (Van Zaalen et al., 2009b; Ward, 2006). Research showed a high frequency of word and phrase repetitions in PWC (Van Zaalen et al., 2009c). Van Zaalen et al., (2009b) suggested that the language disturbances in cluttering appear in order to gain time to complete the editing phase of sentence structuring. The most important reason for this 'time extension' suggestion is the comparison of sentence structure between PWC and controls. PWC produced correct sentence structures comparable to controls when looked at the structures after removal of word and phrase repetitions, interjections and revisions. It was assumed that the word or phrase repetitions extended the editing phase eventually resulting in correct sentence structures. Ward (2004) confirmed this view when he described the language production of adolescents who clutter. Other evidence of this 'time extension' suggestion can be found in the difference between spoken and written language. Where PWC appear to make sentence structure errors in running speech, written sentences are often correct, especially when written at a slow rate (Van Zaalen & Winkelman, 2009). That means that a PWC is capable of producing correct sentences when there is enough time. When a system is not automated time extension will provide possibilities to detect and repair mistakes and complete a process accurately.

The articulator and cluttering

Speech intelligibility reflects the percentage of speech units understood correctly by a listener in a communicative situation. Problems of the articulator in cluttering are mostly related to intelligibility. Indication of automatisation problems at the level of the articulator (execution of planned speech) were not found in this study. Ward (2006) describes "*a reduced ability to maintain articulatory accuracy at higher speech rates"* (Ward, 2006, p.143) as indicative of articulator problems in cluttering. It is argued that in fact these errors are a result of problems in dealing with phonological encoding rather than a result of problems in timing of muscle movements. Some indications of unaffected motor movements of PWC was provided by Hartinger and Mooshammer (2008) who studied the spatial and temporal kinematic variability by means of electromagnetic midsagittal articulography (EMMA) of PWC and PWS. The results showed a significantly higher coefficient of variation for PWC in long words with a complex syllable structure whereas the means of displacements and durations did not differ between PWS and PWC. Hartinger and Mooshammer (2008) and Van Zaalen et al., (2009a) observed that PWC improve their intelligibility by concentrating on their speech task. They concluded that the intelligibility of PWC in a simple task (repeating nonsense syllables) was good, while the intelligibility and articulatory variation in multisyllable words was negatively affected by coarticulation and syllable sequencing errors. It seems reasonable to speculate that articulation (accuracy) in PWC suffers from insufficient resources in phonological encoding.

Subtyping cluttering: syntactical and phonological cluttering

The defective language automatisation hypothesis provides an explanation for the incidences of errors and disfluencies in cluttering, but an explanation is also needed why some PWC produce a high frequency of normal disfluencies at a fast and/or irregular rate, while others produce a high number of speech errors (errors in word or sentence structure or pausing) in the same condition. In the next paragraph on subtyping in cluttering this point will be elaborated in relation to language automatisation and error detection.

Several ideas on subtyping cluttering were presented. Grewel (1970), presented a detailed description of two subtypes of cluttering: the “motor” type and the “sensory” type. Descriptors of the "motor" type of cluttering included: problems in word structure, prosody, rate and rhythm. The "sensory" type of cluttering included, amongst others, lack of fluency in language output. The "sensory" PWC, according to Grewel, did not necessarily exhibit tachylalia. Rather, they speak too fast relative to their own ability, even if language formulation is slow. In 1984, Damsté described three subtypes of cluttering: dysrythmic cluttering, dysartric cluttering and dysphasic cluttering. This proposal was revised by Ward (2006, p. 144), who concluded then two types of cluttering: ‘linguistic cluttering’, similar to Damsté’s dysphasic cluttering and ‘motoric cluttering’, comparable to Damsté’s dysarthric and dysrhythmic cluttering. Based on the collected data in this study and clinical experience subtyping in cluttering seems legitimate.

Syntactic cluttering
In syntactic cluttering, defective automatisation of lexical (lemma) retrieval and grammatical encoding problems can result in word and phrase repetitions, interjections, hesitations and revisions.
Example:
“Mama, I went to the, I went to the library to buy, borrow a book on … what’s its name?’ …. on dinosaurs”.
(Mama I went to the library to borrow a book on dinosaurs)
Phonological cluttering
In phonological cluttering, errors in phonological encoding are not detected by the monitor, resulting in word structure errors (‘motoric effects’: coarticulation, telescoping or syllable sequencing errors) in multi-syllabic words.
Example:
“*Mama, wentlibri to browabook on dinsor*” (coarticulation/telescoping)
(Mama I went to the library to borrow a book on dinosaurs)

Table.4: Description of syntactical and phonological cluttering.

A subgroup of PWC produces excessive hesitations in language production due to lexical retrieval and grammatical encoding problems (interjections, revisions, hesitations, word- and phrase repetitions), where another subgroup produces more errors due to phonological encoding problems (word structure errors). Contrary to Ward’s suggestion of ‘*linguistic and* *motoric cluttering*', a psycholinguistically motivated classification of cluttering is suggested: syntactic and phonological cluttering. Syntactic cluttering (as described in Table 4.) is similar to Ward's description of 'linguistic cluttering', and 'phonological cluttering' is comparable to 'motoric cluttering'. The difference between Ward's classification and the presented classification in this study is mainly based on terminology. The term 'linguistic' refers to "*disorganized and confused language production with maze behaviour*" (Ward, p.144). It is assumed that the term 'syntactic cluttering' narrows the formulation of the observed problems in grammatical encoding in this subtype of cluttering. Contrary to Ward's description of motor cluttering, problems in motor execution were not observed in this study. The observed high incidences of problems in syllable flow and sequencing together with fast rushes of speech in a subgroup of cluttering adults seems to indicate problems in phonological encoding rather than problems in motor execution. In this phonological cluttering, articulation suffers from insufficient attentional resources for phonological encoding.

Summarizing, it can be assumed that cluttering is a nosological entity, distinguishable from stuttering and other speech production problems. Persons who clutter (PWC) can be differentiated from persons who stutter (PWS) by speech rate in retelling a story or producing strings of non sense syllables. Another way to differentiate PWC from PWS is by accuracy, flow and sequencing errors in repeating low frequency multi syllabic words at a fast rate. PWC and PWS also show other levels of brain activations during fMRI speech at a fast rate. PWC can be differentiated from LD-children by the relative frequency of word- and sentence repetitions and the amount of correct sentence structures in retelling a memorized story.

Cluttering, stuttering and speech impairment related to learning disability seem to have different underlying neurolinguistic processes. Children with speech impairment related to learning disability seem to be disfluent as a result of problems in conceptualising. According to Wijnen (2000) in stuttering excessive and lopsidedly focussed monitoring is detrimental, rather than beneficial to speech fluency, as it engenders many unnecessary interruptions and restarts of the ongoing speech. PWC seem to have a 'double deficit' of language automatisation: a deficit in encoding (grammatical and phonological) and as a result of that, weak monitoring, because processing capacity is used for the encoding processes. Further research on the role of automatisation in forming and monitoring the preverbal message, language formulation and speech production is needed in order to confirm this assumption.

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Summary

Summary

This study had the two following objectives: (1) clinical, diagnostic classification of the syndrome of cluttering and particularly the difference in symptomatology between cluttering and stuttering and between cluttering and speaking problems associated with learning disability; (2) to contribute to a (neurolinguistic) model of cluttering that provides a coherent explanation for the observed symptomatology; and elucidates the difference between cluttering and stuttering. The first part of this thesis (Chapter 2, 3, 4 and 5) focuses on evaluation of speech characteristics of persons with fluency disorders. The second part (Chapter 6 and 7) focuses on the underlying neurolinguistic processes in cluttering.

Part I. Differential diagnostics on cluttering

Speech-language pathologists generally agree that cluttering and stuttering represent two different fluency disorders. Differentiating cluttering and stuttering is difficult because these disorders have similar characteristics and often occur in conjunction with each other. Therefore, the first chapter of part I (Chapter 2) describes an empirical study aiming to set objective norms for differential diagnostic assessment of cluttering and stuttering symptoms, based on the three main characteristics of cluttering indicated by St. Louis, Raphael, Myers & Bakker (2003). According to the St. Louis et al., (2003) working definition of cluttering a high and/or irregular articulatory rate is a main characteristic in differential diagnostics between cluttering and stuttering. However, agreement on what defines abnormally fast and abnormally irregular articulatory rate was needed. Participants in this study had been referred to centres for stuttering therapy with self-reported fluency problems. Participants were diagnosed based on (subjective) clinical assessment by two speech language pathologists specialized in fluency disorders. They performed three different speaking tasks: speaking spontaneously, reading aloud, and retelling a story. The objective measures on mean articulatory rate (MAR), accuracy, smooth-flow and type and frequency of disfluencies are compared to the subjective clinical judgment made by fluency experts.

The most striking result of this study was that the correlation between SLPs subjective diagnoses was low ($r = .638$). Furthermore, the MAR of persons who stutter (PWS) was lower compared to that of persons who clutter (PWC). Not all persons diagnosed with cluttering had an abnormally high mean articulatory rate. PWC produced 6.4 - 7.6 times more normal (non-stutter-like) disfluencies compared to stutter-like

disfluencies in spontaneous speech and retelling a memorized story. PWC produced significantly more phonological errors compared to controls and PWS in repeating multi syllabic word strings.

Adding objective measurements to the subjective clinical judgment appeared to be of substantial diagnostic value, especially in locating a cluttering component in speech or language characteristics (based on the characteristics described in the St. Louis et al., (2003) working definition of cluttering). The objective measurement values in this study, described in Chapter 2, are based on a small group of disfluent participants that both SLPs agreed upon. Future studies on cluttering and stuttering should include measurements of multiple factors (speech and language production and fluency) in the data collection process in order to be of substantial diagnostic value.

Another result of this study was that although considerable individual differences between PWC were observed, the group as a whole exhibited language problems to some extent similar to those described in LD-children. Consequently, a systematic comparison between LD-children, CWC and CWS was set up, which made use of the Predictive Cluttering Inventory (Daly and Cantrell, 2006). Chapter 3 discusses the results of these three groups on the Dutch translation of the Predictive Cluttering Inventory (PCI) (Van Zaalen & Winkelman, 2009) in connection to the subjective and objective measurements studied in Chapter 2. In this study 137 Dutch speaking participants ranging in age between 10.6 – 12.11 years, were tested with the PCI by eight different SLPs. Classification was based on SLPs diagnosis as described in Chapter 2. The Predictive Cluttering Inventory was completed on the basis of observation of spontaneous speech, retelling a memorized story, reading aloud, and parental information. Pearson correlations were used to determine relationships between subjective and objective clinical judgment to the checklist norm studied. It was found that the Predictive Cluttering Inventory was not sensitive and specific enough to detect cluttering. A factor analysis was constructed to determine factors that may explain the variance present in the basic variables: speech production, language production, alertness and other behaviours. Results of the factor analysis were compared to results from a cluster analysis. Based on the significantly different items a revised PCI was conducted. The interpretation of item scores in the revised PCI heightened the sensitivity to a low but acceptable score of 69%. (A sensitivity score of 70% is considered acceptable.) Due to the fact that some items and issues concerning the scoring system are not absolutely clear to all SLPs a short manual with clarification of item content and the scoring system could further heighten the sensitivity of the PCI.

The findings described in Chapter 2 and 3 provided ideas for differential diagnosis and set normative objective data on articulatory rate, disfluencies and speech behaviour correlated to cluttering. Preliminary results on the SPA-test (Screening Pittige Articulatie) in a small group of PWC (see Chapter 2) made clear that an extensive study, involving larger groups of participants, and specifically addressing speech motor control at word level in cluttering was needed to improve the differential diagnosis. Chapter 4 describes the study in which the validity of the SPA-test, specifically designed to assess speech motor control at the word level, was tested. The focus of this study was the question if cluttering is a fluency disorder in which speech motor control on word level is disturbed in high speech rate. In an elicitation procedure, repetitions of complex multi-syllabic words at a fast speech rate were obtained from 47 dysfluent participants and 327 controls. Speech production was judged on articulatory accuracy, smooth-flow (coarticulation, flow and sequencing) and articulatory rate. Results from PWC and PWS were compared to normative data based on control group data. PWC produced significantly more accuracy, flow and sequencing errors compared to PWS. Results were interpreted in the way that PWC appeared to have major difficulties in coordinating speech planning and execution at a high articulatory rate. Because speech motor control at word level and phonological encoding are interrelated this study is supportive to the idea that cluttering can be considered to be based on defective language automatisation.

Data of the cluttering group presented in Chapter 2, 3 and 4 also show that within the cluttering group the frequency of word/sentence structure errors as well as disfluencies is highly variable (across individuals). PWC differentiate themselves from PWS by speech characteristics (disfluency ratio, mean articulatory rate and amount of phonological errors). And PWC experience language production disturbances similar to persons with learning disability. In Chapter 5 a study on the extent and type of disturbances in the fluency of language production of CWC and LD-children (LD) was reported. In doing this more knowledge on underlying processes of language production disturbances in these children is gained. In this study 103 Dutch speaking children ranging in age between 10.6 – 12.11 years were divided into three groups. To answer the research question on language disturbances and their underlying neurolinguistic processes production of the following variables were studied: (a) percentage of primary plot elements, secondary plot elements or noise. (b) percentage syntactically correct sentence structures at first or second attempt. (c) percentage sentence structure errors, and (d) the type and frequency of disfluencies.

The results showed that LD-children reproduced fewer primary and secondary plot elements, fewer correct sentence structures at first and second attempt and more incorrect sentence structures compared to children who clutter (CWC) and controls. CWC exhibited story elements, percentage correct sentence structures and sentence structure errors comparable to controls. Although CWC experience language planning disturbances in running speech their level of syntactical correct sentence structures in retelling a memorized story was comparable to fluent controls.

In analysing type, frequency and ratio of disfluencies a difference was found that was not observed earlier. CWC produced significantly more word- and phrase repetitions compared to LD-children and controls. In producing repetitions of already produced words CWC seem to gain time for their formulation process. The high level of self repairs (revisions) and repetitions in CWC and LD-children can support the hypothesis that the automatisation in language formulation is not completed in both CWC and LDC.

Part II: Underlying neurolinguistic processes

In Part II the underlying neurolinguistic processes in cluttering and other disorders of fluency are described and interpreted in the framework of (normal) spoken language production. Chapter 6 reports on a (cognitive) neuroimaging study with the purpose to investigate if cluttering and stuttering are different at the level of the neurological substrate. It was assumed that if different patterns of brain activation are seen in PWC and PWS when performing speech tasks, this would be indicative of differences in underlying neurolinguistic processes / deficits. The participants were carefully screened, in order to only include cluttering individuals without co-occurring stuttering, and stuttering individuals without co-occurring cluttering. The tasks involved speaking words of increasing phonological complexity. A block design with a 40 second on-off period that alternated viewing symbols and reading aloud words with speech was used. Both groups showed a speech related BOLD response predominantly in the frontal lobe and sub-cortical nuclei. Group contrasts indicated that AWC showed higher activity in right hemisphere precentral gyrus, inferior frontal gyrus, and left insula. AWS showed higher activity in the right motor cortex, temporal lobe and globus pallidus. Results on the increase of phonological complexity of the speech tasks were inconclusive concerning the differentiation between

AWS and AWC. Based on the results cluttering and stuttering are different disorders with different underlying activation processes within the central nervous system. Further research in comparison with normal controls is needed to interpret the findings of this study in relation to underlying neurolinguistic processes.

Finally, in Chapter 7 a general discussion of cluttering symptoms within Levelt's (1989) language production model is given. Underlying neurolinguistic processes are described in relation to articulatory rate. In this chapter the question whether cluttering can be seen as a problem in language production automatisation is addressed. Or in other words: Is language output in PWC affected, as a result of attempts at encoding within an insufficiently long time span? It is assumed that fast speech rate reduces the time for conceptualising, formulation and monitoring. When language production is automated, speech rate will have limited effect on speech language production. When language production is to less extent automated, speech rate will have a longer effect on speech language production. It is proposed that in PWC this defective automatisation of language production is exhibited in errors in the conceptualiser, grammatical encoding or phonological encoding and possibly also in the articulator.

Concerning errors in conceptualising (e.g. pragmatic errors, unsatisfactory topic introduction, not adjusting story content to the listener) although documented for cluttering, no conclusive evidence is found in this study. Research on the effect of rate on the conceptualiser is needed to better understand this aspect in cluttering.

Supporting evidence for a defective automatisation in language formulation in cluttering is described in Chapters 4, 5, and 7. In running speech, PWC exhibit more grammatical and phonological encoding errors in comparison to fluent speakers. But, speaking with full attention (retelling a story, slow speech rate) elicits errors and repairs in PWC according to controls. The fact that PWC can endure this attentive speech only for a short time can also be a sign of automatisation problems.

Some evidence for defective automatisation in language production in cluttering for effects on the articulator is described in Chapter 4. In a fast rate PWC exhibit more speech errors (accuracy, flow and sequencing) compared to stuttering and fluent controls. Other supportive data was provided by Hartinger and Mooshammer (2009) who concluded that articulatory variation in cluttering is higher compared to stuttering.

Based on the data of this study, it can be concluded that cluttering is merely a problem of automatisation in language production. Why PWC are not able to adjust their speech rate to these language production problems is not studied in this project. This study has provided a possible explanation why PWC can not adjust their speech rate for a long time. It is possible that a PWC is not able to monitor his speech rate because attentional resources are needed to overcome the defective automatisation of language production. Further research on language production (e.g. grammatical and phonological encoding in spontaneous speech) and brain activation (cerebral dominance and basal ganglia activation) in cluttering can be supportive in understanding the intriguing disorder of cluttering.

Samenvatting

Deze studie had de twee volgende doelstellingen: (1) klinische, diagnostische classificatie van het syndroom broddelen en in het bijzonder het verschil in symptomatologie tussen enerzijds broddelen en stotteren en anderzijds tussen broddelen en spreekmoeilijkheden gerelateerd met leermoeilijkheden; (2) een bijdrage leveren aan een samenhangend (neurolinguïstisch) model van broddelen dat een verklaring geeft voor de geobserveerde symptomatologie, en tegelijkertijd het verschil tussen broddelen en stotteren toelicht.

Het eerste deel van de dissertatie (Hoofdstuk 2, 3, 4 en 5) richt zich op de evaluatie van spraak en taalkenmerken van personen met vloeiendheidsstoornissen. Het tweede deel (Hoofdstuk 6 en 7) richt zich op de beschrijving van de onderliggende neurolinguïstische processen bij broddelen.

Deel I. Differentiaaldiagnostiek bij broddelen

Logopedisten en spraaktaalpathologen zijn het er over het algemeen over eens dat broddelen en stotteren twee verschillende vloeiendheidsstoornissen vertegenwoordigen. Differentiaaldiagnostiek tussen broddelen en stotteren is moeilijk omdat deze stoornissen gelijke kenmerken hebben en vaak in samenhang voorkomen.

Het eerste hoofdstuk van deel I (Hoofdstuk 2) beschrijft een empirische studie om objectieve normen te bepalen ten behoeve van differentiaaldiagnostisch onderzoek bij broddel- en stotterkenmerken op basis van de drie kernkarakteristieken van broddelen geïndiceerd door St. Louis, Raphael, Myers & Bakker (2003). Volgens de St. Louis e.a. (2003) is een hoog en/of onregelmatig spreektempo een kernkarakteristiek van de differentiële diagnose tussen broddelen en stotteren. Echter, overeenstemming over wat een abnormaal snel of abnormaal onregelmatig spreektempo, is noodzakelijk. Proefpersonen in deze studie waren verwezen naar praktijken voor stottertherapie in het centrum van Nederland vanwege zelf gerapporteerde problemen in de vloeiendheid van het spreken. Proefpersonen werden gediagnosticeerd op basis van een (subjectieve) klinische beoordeling door twee logopedisten die zich gespecialiseerd hebben in de diagnostiek en behandeling van cliënten met vloeiendheidsstoornissen. Er werd gebruik gemaakt van opnamen van drie verschillende spreektaken: spontaan spreken, lezen en het navertellen van een verhaal. Objectieve meetresultaten werden verzameld met betrekking tot: articulatietempo, articulatorische nauwkeurigheid, geleidelijkheid (coarticulatie, gelijkmatigheid en opeenvolging) en snelheid, frequentie en type van niet-vloeiendheden.

De objectieve meetresultaten werden vergeleken met de subjectieve klinische beoordelingen gedaan door de gespecialiseerde logopedisten.

Het opvallendste resultaat van de studie was dat de correlatie tussen de subjectieve diagnoses erg laag was (r=.638). Verder bleek dat de gemiddelde articulatiesnelheid van de personen die stotteren (PDS) lager was vergeleken met personen die broddelen (PDB), zoals werd voorspeld. Niet alle broddelende proefpersonen hadden een hoog gemiddelde articulatiesnelheid vergeleken met controles. PDB produceerden 6.4 – 7.6 keer zoveel normale niet-vloeiendheden in verhouding tot stotter-niet-vloeiendheden in spontane spraak en het navertellen van een verhaal. PDB produceerden significant meer fouten in accuraatheid tijdens de herhaling van meerlettergrepige woordreeksen in vergelijking met controles en PDS.

Toevoeging van objectieve meetresultaten aan de subjectieve klinische beoordeling bleek van substantiële diagnostische waarde, in het bijzonder in het lokaliseren van een broddelcomponent (gebaseerd op karakteristieken beschreven in St. Louis et al., 2003 werkdefinitie van broddelen). De objectieve meetresultaten in deze studie, beschreven in hoofdstuk 2, zijn gebaseerd op een kleine groep niet-vloeiende proefpersonen van wie de gespecialiseerde logopedisten onafhankelijk van elkaar een overeenstemmende diagnose gesteld hadden. Verdere studies naar broddelen en stotteren zouden meerdere factoren en domeinen in de dataverzameling moeten omvatten om van substantieel diagnostische waarde te zijn.

Een ander resultaat van de studie was de ontdekking dat alhoewel er aanzienlijke individuele verschillen tussen de broddelaars waren, de groep als geheel taalproblemen vertoonde die vergelijkbaar zijn met de taalproblemen die toegeschreven worden aan kinderen met leermoeilijkheden. Dientengevolge werd een systematische vergelijking tussen kinderen met leermoeilijkheden, kinderen die broddelen, kinderen die stotteren en controles opgezet, die gebruik maakte van de Predictive Cluttering Inventory (Daly and Cantrell, 2006). In hoofdstuk 3 worden resultaten van de Nederlandse vertaling van de PCI (Checklist Broddel Kenmerken, Van Zaalen & Winkelman, 2009) besproken in relatie tot de subjectieve en objectieve meetresultaten besproken in Hoofdstuk 2. In deze studie werden 137 Nederlands sprekende kinderen variërend in leeftijd van 10.6 – 12.11 jaar, gescreend met de PCI door 8 verschillende logopedisten. De classificatie was gebaseerd op de diagnose van de gespecialiseerde logopedisten zoals beschreven in hoofdstuk 2. De leeftijdsvariatie in alle groepen was gelimiteerd om bias tengevolge van ontwikkelingsinvloeden te beperken. De PCI werd ingevuld op basis van de observatie van spontane spraak, navertellen van een verhaal, lezen en ouderinformatie. Pearson's correlaties werden gebruikt om de relatie tussen enerzijds de subjectieve klinische beoordeling aangevuld met objectieve metingen en anderzijds de score op de checklist te beoordelen. De PCI bleek niet sensitief of specifiek genoeg om broddelen te detecteren. Een factoranalyse werd uitgevoerd om de factoren te bepalen die gezamenlijk de variantie in de basisvariabelen kunnen verklaren. Resultaten van de factoranalyse werden vergeleken met resultaten van een clusteranalyse. De interpretatie van de itemscores van de gereviseerde PCI zorgde voor een verhoging van de sensitiviteit tot een lage, maar acceptabele waarde van 69%. (Een sensitiviteitsscore van 70% is acceptabel). Aangezien sommige onderwerpen en enkele zaken betreffende het scoringssysteem nog niet volledig duidelijk waren voor alle logopedisten, wordt verondersteld dat een korte handleiding met toelichting op de iteminhoud de sensitiviteit van de PCI-r verder kan verhogen. De ondersteunende, maar niet significant verschillende, symptomen op de lijst kunnen van groot belang zijn voor de planning van de therapie voor de individuele cliënt.

De studies beschreven in hoofdstuk 2 en 3 hebben ideeën voor differentiaaldiagnostiek en normatieve waarden voor articulatiesnelheid en variatie, niet-vloeiendheden en ander aan broddelen gerelateerd spreekgedrag opgeleverd. Voorlopige resultaten van de SPA -test (Screening Pittige Articulatie) in een kleine groep PDB (zie hoofdstuk 2) maakten duidelijk dat uitgebreider onderzoek (grotere groep proefpersonen) naar spraakmotorische controle op woordniveau in broddelen een bijdrage kan leveren aan de differentiaaldiagnose. Hoofdstuk 4 beschrijft een studie waarin de validiteit van de SPA-test, speciaal ontworpen om spraakmotorische controle op woordniveau te onderzoeken, werd getest. Speerpunt in deze studie was de vraag of broddelen een vloeiendheidstoornis is waarin de spraakmotorische controle (of mogelijk de fonologische codering) verstoord is binnen een hoge spreeksnelheid. Er werd verondersteld dat een verstoorde spraakmotorische controle zou resulteren in fouten in accuraatheid, gelijkmatigheid van het spreken en de opeenvolging van lettergrepen. Binnen een vaststaande procedure werden op hoge snelheid herhalingen van meerlettergrepige woorden uitgelokt bij 47 niet-vloeiende sprekers en 327 controles. Spraakproductie werd beoordeeld op articulatorische accuraatheid, geleidelijkheid en snelheid. Resultaten van PDB en PDS werden vergeleken met de normatieve data verkregen van de controles. In vergelijking met PDS produceerden PDB significant meer accuraatheid, gelijkmatigheid en opeenvolgingfouten. Uit de resultaten werd geïnterpreteerd dat PDB aanmerkelijke problemen ervaren in spraakmotorische planning en uitvoer binnen een hoog spreektempo. In PDB werden veel verstoringen van de planning waargenomen; dit impliceert mogelijk dat de taalproductie niet volledig was geautomatiseerd. Aangezien spraakmotorische controle op woordniveau en fonologische codering nauw met elkaar verbonden zijn, ondersteunen de resultaten uit deze studie het idee dat broddelen gezien kan worden als een aan taalproductie gerelateerde vloeiendheidstoornis.

Data van de broddelgroep gepresenteerd in hoofdstuk 2, 3 en 4 laten ook zien dat binnen de broddelgroep een grote marge van woord/zinsstructuurfouten en frequentie van normale niet-vloeiendheden bestaat. PDB onderscheiden zich van PDS op spraakkarakteristieken (ratio niet-vloeiendheden, gemiddeld articulatietempo en aantal fonologische fouten). Differentiaaldiagnostische kenmerken onderscheiden een stoornis van alle andere stoornissen. PDB ervaren taalproductieverstoringen die vergelijkbaar zijn met die van personen met leermoeilijkheden (LM). In hoofdstuk 5 is een studie naar het type en de hoeveelheid taalproductieverstoringen van kinderen die broddelen (KDB) en KLM beschreven. Op deze manier werd meer kennis aangaande de onderliggende processen van de taalproductie van deze kinderen verworven. In deze studie werden 103 Nederlands sprekende kinderen variërend in leeftijd van 10.6-12.11 jaar verdeeld over drie groepen (broddelen, leermoeilijkheden en controles). Om de vraag te beantwoorden aangaande de taalproductieverstoringen en de onderliggende neurolinguïstische processen werden de volgende variabelen bestudeerd tijdens het navertellen van een verhaal: (a) percentage hoofd- en bijzaken en ruis; (b) percentage syntactisch correcte zinsstructuren zonder en met zelfcorrectie; (c) percentage zinnen met syntactische fouten en (d) percentage en type niet-vloeiendheden.

Resultaten toonden aan dat KLM minder hoofd- en bijzaken navertelden, minder correcte zinstructuren (zonder en met zelfcorrectie) en meer zinnen met grammaticale fouten produceerden. KDB produceerden een aan de controles vergelijkbare hoeveelheid hoofd- en bijzaken en correcte zinsstructuren. Dus, hoewel het idee bestaat dat KDB in spontane spraak een meer dan normale hoeveelheid taalproductieverstoringen vertonen, is hun taalproductie in het navertellen van een verhaal vergelijkbaar met die van vloeiend sprekende controles.

Bij het analyseren van het type, de frequentie en de ratio van de niet-vloeiendheden werd een niet eerder beschreven onderscheid tussen KDB en KLM ontdekt. KDB produceerden een significant grotere hoeveelheid woord- en zinsdeelherhalingen in vergelijking met KLM en controles. Er is verondersteld dat KDB in het produceren van de herhalingen van al geproduceerde woorden of zinsdelen tijd winnen voor het proces van taalformulering. Het hoge aantal zelfcorrecties in KDB en KLM ondersteunt de gedachte dat zowel bij KDB als KLM de automatisering van de taalproductie nog niet volledig is.

Deel II: Onderliggende neurolinguïstische processen

In deel II worden de onderliggende neurolinguïstische processen bij broddelen en andere vloeiendheidsstoornissen beschreven en geplaatst binnen een model van taalproductie. Hoofdstuk 6 beschrijft een neurocognitieve beeldvormende studie met het doel om te onderzoeken of broddelen en stotteren verschillen op het niveau van neurologische substraat. Er werd verondersteld dat als verschillende patronen van hersenactiviteit gezien werden tijdens de uitvoer van spreektaken bij volwassenen die broddelen (VDB) en volwassenen die stotteren (VDS), dit indicatief is voor verschillen in onderliggende neurolinguïstische processen of defecten. De participanten werden nauwkeurige gescreend, om PDB te rekruteren zonder bijkomende stotterkenmerken en PDS zonder bijkomende broddelkenmerken. De spreektaak bestond uit het herhaald opzeggen van woorden met toenemende fonologische complexiteit. In een blokdesign met een 40 sec aan-uit periode werden afwisselend symbolen gelezen en gelezen woorden hardop uitgesproken. Beide groepen vertoonden voornamelijk aan spraak gerelateerde hersenactiviteit in de frontale lobus en de sub-corticale nuclei. Groepsvergelijkingen gaven aan dat VDB ten opzichte van VDS een hogere activiteit hadden in de precentrale gyrus en de inferior frontale gyrus van de rechterhemisfeer en de linker insular cortex. PDS vertoonden een hogere activiteit in de rechter motor cortex, temporale lobus en de globus pallidus. Gebaseerd op deze bevindingen kan verondersteld worden dat broddelen en stotteren verschillende stoornissen zijn met andere onderliggende neurolinguïstische processen.

Afsluitend werden in een algemene discussie de broddelkenmerken in verband gebracht met het taalproductiemodel van Levelt (1989). Onderliggende neurolinguïstische processen bij broddelen zijn beschreven in relatie tot articulatietempo. In deze generale discussie in hoofdstuk 7 werd de vraag beantwoord in hoeverre broddelen gezien kan worden als een uiting van onvolkomen automatisering van de taalproductie. Met andere woorden: Is de talige output van PDB beïnvloed als resultaat van een poging om te encoderen binnen een ontoereikende tijdsspanne? Er wordt verondersteld dat een hoog spreektempo de tijd voor de conceptualiser, de formulator en de monitoring reduceert. Als de taalproductie volledig geautomatiseerd is zal het spreektempo nauwelijks invloed uitoefenen op de spraak- en taalproductie. Indien taalproductie in mindere mate geautomatiseerd is, zal verandering van spreektempo een effect van groter belang hebben op de spraak taal productie. Er wordt verondersteld dat een onvolkomen automatisering van de taalproductie in PDB zich uit in fouten in de conceptualiser, grammaticale en/of fonologische codering en mogelijk ook in de articulator.

Ten aanzien van fouten in de conceptualisator (bijv. pragmatische fouten, onvoldoende onderwerpintroductie, het niet aanpassen van de verhaalinhoud aan de luisteraar) werden, alhoewel gedocumenteerd voor broddelen, binnen deze studie geen sluitende bewijzen gevonden. Verder onderzoek naar het effect van spreeksnelheid en automatisering op de accuraatheid van de conceptualisator is nodig om dit aspect binnen broddelen beter te begrijpen.

Ondersteunend bewijs voor een onvolkomen automatisering van taalformulering (de formulator) is beschreven in de hoofdstukken 4, 5 en 7. In spontane spraak produceren PDB meer grammaticale en fonologische coderingsfouten in vergelijking met controles. Maar, tijdens het spreken met aandacht voor het spreekproces (navertellen van een verhaal, spreken in een langzamer tempo) werden bij PDB fouten en zelfcorrecties waargenomen die in soort en aantal vergelijkbaar zijn met die van vloeiende sprekers. Het feit dat PDB deze vorm van aandachtig spreken slechts gedurende korte tijd kunnen volhouden mag ook gezien worden als een teken van een onvolkomen automatisering van de taalproductie.

Enig bewijs voor een onvolkomen automatisering van de taalproductie op het gebied van de articulator is voor broddelen beschreven in hoofdstuk 4. In een snel tempo produceert de PDB meer spreekfouten (accuraatheid, gelijkmatigheid en opeenvolging) vergeleken met PDS en controles. Ander ondersteunend bewijs werd geleverd door Hartinger en Mooshammer (2007) die concludeerden dat de variatie in articulatiepositie in PDB hoger is dan bij PDS.

Gebaseerd op de resultaten van dit onderzoek mag geconcludeerd dat broddelen voornamelijk een uiting is van een onvolkomen automatisering van de taalproductie. Waarom PDB niet in staat zijn om hun spreektempo aan te passen is binnen deze studie niet onderzocht. Dit onderzoek heeft een mogelijke verklaring opgeleverd voor de problemen van PDB om hun spreektempo gedurende langere tijd aan te passen. Het kan zo zijn dat het voor een PDB door een onvolkomen automatisering van de taalproductie niet mogelijk is om naast de extra aandacht die gegeven moet worden aan de taalproductie nog extra aandacht te besteden aan het monitoren van het spreken (de verlaging van de spreeksnelheid). Verder onderzoek naar de taalproductie (bijvoorbeeld grammaticale en fonologische codering in spontaan spreken) en hersenactiviteit (bijvoorbeeld cerebrale dominantie en activiteit van de basale ganglia) bij broddelen tijdens spreektaken kan ondersteunend zijn in het vergaren van meer kennis over deze intrigerende stoornis.

Abbreviation list

AWC	Adult(s) who clutter
AWS	Adult(s) who stutter
CSSf	Correct sentence structure first attempt
CSSs	Correct sentence structure second attempt
CWC	Children who clutter
CWS	Children who stutter
fMRI	Functional Magnetic Resonance Imaging
GAS	Gemiddelde articulatiesnelheid
KDB	Kinderen die broddelen
KLM	Kinderen met leermoeilijkheden
LD	Learning disability
LM	Leermoeilijkheden
MAR	Mean Articulatory Rate
NDF	Percentage normal disfluencies
OMAS	Oral Motor Assessment Screening (Riley, 1984)
PCI	Predictive Cluttering Inventory (Daly & Cantrell, 2006)
PCI-r	Revised Predictive Cluttering Inventory (Van Zaalen & Winkelman, 2009)
PDB	Perso(o)n(en) die broddelen
PDS	Perso(o)n(en) die stotteren
PPE	Primary plot element
PWC	Person(s) who clutter
PWCS	Person(s) who clutter-stutter
PWS	Person(s) who stutter
RD	Ratio disfluencies
SD	Standard deviation
SLP	Speech language pathologist
SPA	Screening Pittige Articulatie (Van Zaalen & Winkelman, 2009)
SPE	Secondary plot element
SSE	Sentence structure errors
SSI-3	Stuttering Severity Instrument-3 (Riley & Riley, 1994)
VDB	Volwassene(n) die broddelen
VDS	Volwassene(n) die stotteren

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Curriculum vitae

Yvonne van Zaalen-op ’t Hof is in 1966 geboren in Haarlem. In 1984 behaalde zij in Beverwijk haar VWO diploma. Haar HBO diploma logopedie behaalde zij in 1988 in Utrecht. Inmiddels is Yvonne meer dan twintig jaar werkzaam als logopedist, en heeft zij zich gespecialiseerd in vloeiendheidsstoornissen. In 1994 behaalde zij haar HBO master opleiding stottertherapie in Rotterdam. Sinds 2006 is zij tevens werkzaam op de Fontys Hogeschool te Eindhoven, waar zij naast haar werk als docent vloeiendheidsstoornissen en evidence based practice, vanuit de kenniskring van het lectoraat verantwoordelijk is voor de coördinatie van toegepast wetenschappelijk onderzoek binnen de paramedische hogeschool. In 2006 behaalde zij haar titel master of science in de Logopediewetenschap. Sinds mei 2007 is Yvonne de eerste ‘internationale coördinator van klinische zaken’ van de International Cluttering Association. Yvonne geeft post-HBO cursussen op het gebied van zowel stotteren als broddelen en is een veel gevraagde spreker op nationale en internationale congressen. Meerdere artikelen op haar naam zijn verschenen in internationale peer-reviewed tijdschriften. In mei 2009 is het boek “*Broddelen, een (on)begrepen stoornis*”, van haar hand verschenen bij uitgeverij Coutinho. Dit boek dat Yvonne geschreven heeft in samenwerking met Coen Winkelman beschrijft de historie en etiologie van broddelen, biedt een nauwkeurig kader voor zowel de onderkennende, verklarende en ook de handelingsgerichte diagnostiek en goed gemotiveerde aan de praktijk getoetste handreikingen voor therapie.