

SIG 4

Research Article

An Adolescent Confronted With Cluttering: The Story of Johan

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ABSTRACT

Purpose: This case study is presented to inform the reader of potential speech, language, cognitive, and emotional characteristics in preadolescent cluttering. **Method:** This case study describes a 10-year-old boy who started to clutter during preadolescence. The case illustrates that, in some adolescents, cluttering can co-occur with temporary stuttering-like behavior. In this case, signs of disturbances in speech-language production associated with behavioral impulsiveness as a young child were noted. Speech, language, cognitive, and emotional results of the case are reported in detail.

Results: The changes in fluency development are reported and discussed within the context of changes in the adolescent brain as well as adolescent cognitive and emotional development. While being unaware of their speech condition before adolescence, during preadolescence, the changes in brain organization lead to an increase in rate and a decrease in speech control. Given that the client had limited understanding of what was occurring, they were at risk of developing negative communication attitudes. Speech-language therapists are strongly advised to monitor children with cluttering signals in the early years of their adolescence.

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Cluttering is a communication disorder in which a person is not able to adjust their articulatory rate to the linguistic or motor demands of the moment (van Zaalen, 2022), resulting in errors in pausing together with either a high frequency of normal disfluencies or unintelligible words (St. Louis & Schulte, 2011). During school years and especially in preadolescence and adolescence, a fast increase in articulatory rate can be observed. Studies of articulatory rate have been conducted following the same protocol. The mean articulatory rate (MAR) in conversations of fluent Dutch children is measured between 3.3 syllables per second (SPS; Boey, 2003) and 4.43 SPS in Hebrew speakers (Amir & Grinfeld, 2011). Articulatory rates increase from 4.45–5.19 SPS at the age of 7 years to

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5.58–5.89 SPS at the age of 9 years. Further increase is seen from the age of 11 years (5.92 SPS) to ages 13 years (7.19 SPS) and 17 years (7.72 SPS; Amir & Grinfeld, 2011). Van Zaalen and Winkelman (2014) found that the MAR declines to 4.8 SPS after the age of 22 years.

Cluttering may not be diagnosed until adolescence or young adulthood, which may be related to this increase in articulatory rate (van Zaalen & Reichel, 2015). Mensink-Ypma (1990) observed that cluttering manifests itself only when language development is in an advanced stage and a person has a strong inner urge to speak. Cluttering can therefore be difficult to identify in children below 10 years of age. There are two potential reasons for this late identification of cluttering. First, the children's speech rate is still too low to have a significant influence on the fluency and intelligibility of their speech production. Second, it is difficult to differentiate whether errors in sentence structures are based on a disorder in language development or only on an insufficient adjustment to rate.

In this article, we present the case of a 10- to 11-year-old boy who started to clutter during preadolescence. This case illustrates the fact that cluttering in some children can only be observed when they reach the early stages of adolescence. This pattern has been found to be especially true for those children who show signs of disturbances in speech-language production associated with behavioral impulsiveness as a young child (Mensink-Ypma, 1990; Weiss, 1964, 1968).

Damste (1990) discussed the presence of subtypes of cluttering. In this article, we refer to the subtyping as described by van Zaalen (2009), van Zaalen & Reichel (2014), van Zaalen (2022), and Ward (2018). Syntactic cluttering refers to problems in grammatical encoding and word retrieval at a fast speech rate. The problems are manifested by normal disfluencies, such as word and phrase repetitions, interjections, hesitations, and revisions (van Zaalen, 2009), for example, "I am am very busy wor-working eh on my paper eh thesis" instead of "I am very busy working on my thesis." Phonological cluttering refers to problems in phonological encoding and is characterized by word structure errors (e.g., coalescence, telescoping, or syllable sequencing errors) at a fast speech rate, especially in multisyllabic words. Ward (2018) refers to this subtype of cluttering as motoric cluttering, for example, "Probly we will teetmorrow" instead of "Probably we will meet tomorrow." Although Ward (2018) and van Zaalen (2022) describe the same symptoms and subtypes, more research is needed to determine if the affected underlying mechanisms are motorically or phonologically based. Similar to syntactic cluttering, symptoms of phonological cluttering occur more frequently when language formulation is compromised (van Zaalen et al., 2008).

Prevalence and Onset of Cluttering

The prevalence of cluttering has not been researched to the point of being conclusive. Literature used to indicate that cluttering is less prevalent than stuttering (Daly & Burnett, 1999; St. Louis et al., 2003). However, recent studies, conducted using the working definition of cluttering in the work of St. Louis et al. (2007) and the causal definition in the work of van Zaalen (2009), have suggested that cluttering (1.1%) is in fact a little more prevalent than stuttering (1.0%) in adolescents and young adults (Schnell et al., 2013; van Zaalen & Reichel, 2017). According to some experts, pure cluttering is present in 5%–16% of the disfluent population (Bakker et al., 2005; St. Louis & McCaffrey, 2005). Prevalence numbers vary within age groups.

Cluttering Onset

A speech-language pathologist may treat children for other communication impairments prior to a diagnosis of cluttering. In our experience, disfluent young children who regain speech control (after therapy) often return for speech therapy when they are between 10 and 13 years of age. In the early years of adolescence, disturbances in communication often reoccur. On the basis of natural development, speech rates tend to become significantly higher when people with cluttering (PWC) use more complex (multisyllabic) words and sentence structures. Their speech becomes less intelligible and less fluent. If cluttering manifests itself in preadolescence, an increase in the severity of symptoms is often noticed in adolescence. We have noted indicators of possible cluttering in early development. The following signs of possible cluttering in young children (< 10 years of age) have been identified by multiple researchers (Damste, 1990; Mensink-Ypma, 1990; van Zaalen & Strangis, 2021; van Zaalen & Winkelman, 2014; Ward, 2018): (a) a fast speech or articulatory rate; (b) telescoping when using multisyllabic words; (c) omitting small words (such as articles and prepositions) when reading aloud, when writing, or during auditory memory tasks; (d) semantic or syntactical errors during fast writing or speech, which disappears in slower writing or speech; (e) relying on guessing while reading, which disappears when auditory feedback is given via a headphone (contrary to dyslexia); (f) errors occurring during reading at a fast rate (but not at a slow rate or when adequately focused); and (g) grapheme substitutions between /b/ and /d/. Mensink-Ypma (1990) and Ward (2006, 2018) stated that cluttering manifests itself when language development is in a far-advanced stage and a person has high urgency to speak. Although these findings are broad and may also be indicative of other difficulties, associations found between these signs and later development of cluttering warrant the monitoring of children with signs of possible cluttering throughout their adolescence. A careful differential diagnosis of cluttering from other difficulties such as learning disorders is also warranted.

In response to Weiss (1964, 1968), Diedrich (1984) observed that the onset of cluttering occurred about 7 years later than the onset of stuttering. This observation is consistent with the finding in several studies that the onset of cluttering occurs in preadolescence (Howell & Davis, 2011; Schnell et al., 2013; van Zaalen, 2009; van Zaalen et al., 2012). Around the time children reach the age of about 10–11 years, their speech rate tends to accelerate. As a result of this natural rate increase, the rate control of adolescents is no longer strong; therefore, in some cases, cluttering characteristics may emerge (van Zaalen & Reichel, 2015; van Zaalen & Winkelman, 2014). Understanding adolescent brain development will help inform understanding the development of cluttering in adolescence.

Adolescent Brain Development

Adolescence is defined as a transitional period between childhood and adulthood characterized by changes

in social interaction and the acquisition of mature cognitive abilities (Caballero et al., 2016). Caballero et al. (2016) state that these changes have been associated with the maturation of brain regions involved in the control of motivation, emotion, and cognition. An accurate conceptualization of cognitive and neurobiological changes during adolescence must treat adolescence as a transitional developmental period (Spear, 2000) rather than as a single snapshot in time (Casey et al., 2005). In other words, to understand this developmental period, transitions into and out of adolescence are necessary for distinguishing distinct attributes of this stage of development (Casey et al., 2008).

The traditional explanation of adolescent behavior has been suggested to be due to the protracted development of the prefrontal cortex (Casey et al., 2008). Casey et al. (2008) take into consideration the development of the prefrontal cortex (which plays a key role in executive functions) together with subcortical limbic regions (e.g., nucleus accumbens) that have been implicated in impulsive choices and actions. The nucleus accumbens is part of the basal ganglia. The basal ganglia network may be viewed as multiple parallel loops and reentering circuits whereby motor, associative, and limbic territories are engaged mainly in the control of movement, behavior, and emotions (Lanciego et al., 2012). The basal ganglia network seems to be involved in (a) the goal-directed system selection and facilitation of prefrontal-striatopallidal activity during the performance and acquisition of new activities and tasks (goal-directed system); (b) reinforcement learning to create habitual responses automatically performed by the motor circuit (habit system); and (c) stopping an ongoing activity and switching to a new one if necessary, which is mainly mediated by the inferior frontal cortex/subthalamic nucleus-cortical circuit (see Lanciego et al., 2012, for a review). In general, abnormalities in these domains and functions lead to movement disorders such as Parkinsonism and dyskinesias, obsessivecompulsive disorders, and alterations of mood (i.e., apathy, euphoria; Lanciego et al., 2012). Their temporary dysfunction in adolescence results in a wide range of neurological conditions, including disorders of behavior control and movement, as well as cognitive deficits that are similar to those that result from damage to the prefrontal cortex (Frank & O'Reilly, 2006).

Cluttering and the Adolescent Brain

The first researchers discussing the role of the basal ganglia in cluttering were Seeman (1970) and Lebrun (1996). On the basis of a functional magnetic resonance imaging study in a group of adult PWC (n = 14) or people with stuttering (n = 16) during repeated production of word and nonword sequences with increased linguistic and motor difficulty, van Zaalen-op't Hof et al. (2009) assumed that cluttering is caused by an inhibition problem

in the basal ganglia. Temporal and sublobar activation of the superior temporal gyrus of the insular cortex, the lentiform nucleus of the putamen, the ventral nucleus of the thalamus of the left- and right-cerebrum precentral gyrus, and the inferior frontal gyrus as well as sublobar extranuclear and temporal activation were significantly higher for PWC compared with people with stuttering (van Zaalen-op't Hof et al., 2009b, p. 114). The results in the work of van Zaalen-op't Hof et al. (2009b) were confirmed by Ward et al. (2015) when they concluded that adults who clutter produced higher activity compared with control speakers in the lateral premotor cortex bilaterally and on the medial surface (pre-Supplementary Motor Area [SMA]). Subcortically, adults who clutter showed greater activity than control speakers in the basal ganglia. Specifically, the caudate nucleus and putamen were overactive in adults who clutter for the comparison of picture description with sentence reading. In addition, adults who clutter had reduced activity relative to control speakers in the lateral anterior cerebellum bilaterally (Ward et al., 2015). Alm (2011) hypothesizes that the hyperactivation and dysregulation of the medial frontal cortex is an underlying mechanism in cluttering. He considered such processes secondary to the disinhibition of the basal ganglia circuits, which can occur, for example, because of a hyperactive dopamine system (Alm, 2011). The SMA proper, along with the basal ganglia and the cerebellum, controls the timing of the articulation and thereby the speech rate. In fluent speakers, the production of speech is monitored on various levels, mainly by auditory connections to the anterior cingulate cortex (ACC) and the SMA (van Zaalen & Reichel, 2015). Functions associated with the ACC and the SMA are as follows: "(1) Drive, motivation, and initiation of action; (2) Inhibition of impulses; (3) Attention; monitoring and correction of behaviour; (4) Planning of sequential behaviour; (5) Selection of words and word-forms; (6) Execution and timing of sequential behaviour" (Alm, 2011, p. 21).

Social-Emotional Component of Cluttering

The social–emotional component concerns the experiences a person with cluttering has accumulated over the years. This means that if a person with cluttering is often misunderstood, fear of communication can develop (van Zaalen & Winkelman, 2014). A person is often unaware of this process, which makes it a hidden problem. The fear starts when a person is frequently not taken seriously because the speaker's message is not understood and no relationship is formed between the response of the listener and the speaker's own speech (Wilhelm, 2020). Unlike in stuttering, fear of specific sounds or words is less likely to develop in pure cluttering.

Cluttered speech seems to increase in circumstances where the person is relaxed (e.g., within the family or with

friends) and has less focus on speech monitoring. The effects that abnormal speech has on the listener reflect in the interpersonal and social component. A high frequency of normal disfluencies and fillers, poor speech intelligibility, and a fast rate of communicative exchange (too short pauses) between the speaker and the listener interfere with the listener's ability to adjust to the speaker. Although the message is clear in the mind of the speaker, the many disruptions within a fast rate and the lack of pauses to process the communicative exchange negatively influence the listener's understanding. The messages of PWC are therefore often misunderstood.

In preadolescence and adolescence, being part of a social group of peers is of great importance. Feeling different because of your own characteristics (e.g., stuttering, cluttering) can determine your social relations to the group. For instance, if peers start to realize that a mate who clutters is sometimes difficult to understand due to their speech, they could start to tease them for the way they speak, ignore them, and/or not give them credit for what is said. The same or other things could happen in a school setting, with teachers assuming that their maze behavior is due to a "lack of study" or a "lack of motivation." In a family context, members may assume that "he is not practicing enough" or "he is not a good communicator" due to his difficult-to-follow or difficult-to-understand speech. As it happens for stuttering (see Boyle, 2013, for a review), we are aware that stereotypes, prejudices, discrimination, and public stigmas from the environments the person who clutters lives in could slowly become self-stigmas (Reichel & St. Louis, 2007), the same being internalized by the person themself. For example, self-stigmas may make PWC believe that they are not a good communicator, thereby changing their self-perception of their role in society and affecting the choices they make in life (e.g., education, social life, work life). For these reasons, in order to avoid these negative social-cognitive-emotional consequences, early treatment is recommended as soon as a negative communication attitude arises (Damste, 1990; Reichel & St. Louis, 2007; van Zaalen & Winkelman, 2014; Winkelman, 1990).

Disfluencies in the Language Production Process

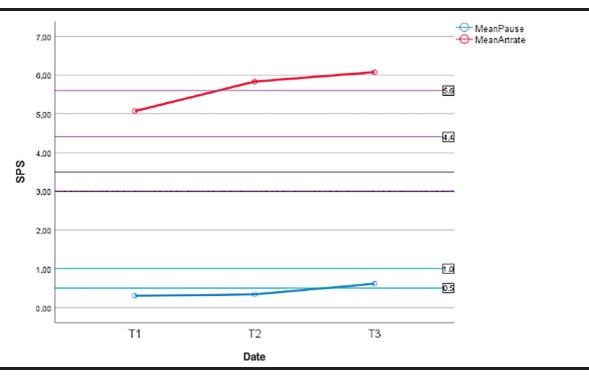
Correct fluent sentences can be produced at a fast speech rate if the process of language formulation is synchronous. Sentences are constructed using appropriate words that are planned and produced as intended. When people clutter, some hypothesize that their language formulation processes are not in synchrony with language production (Myers, 2014). The sentence or word structure is not planned or completed within the available time. When language formulation is insufficiently synchronous, errors in language production can be expected, manifested

by an excessive number of disfluencies or speech errors (van Zaalen & Reichel, 2015). The appearance of normal disfluencies can be best explained by a time-gaining effect (Howell & Au-Yeung, 2002b). A person repeats that part of the message that has already been planned or adds a pause and, by doing so, gains time to plan the rest of the sentence. It is as if the listener can hear the person think and formulate. This behavior is known as maze behavior (Howell & Au-Yeung, 2002b). Mazing is very difficult to follow by the listener because it taxes listener memory a lot. In cluttering, the combination of a fast speech rate, short pauses, disfluencies, and excessive coarticulation makes the perception of disfluencies even higher than the actual number of disfluencies when measured (van Heeswijk, 2012).

Phonological encoding errors occur because they are not detected by PWC when their speech rate is fast (van Zaalen, 2009). Myers (2014) posits that sustained monitoring and modulation of speech production requires careful vigilance by PWC. These skills do not come naturally or automatically to them, especially when they speak spontaneously on topics that they find exciting and complex (St. Louis et al., 2007). To explain the basis of the disfluencies and speech intelligibility problems in cluttering, it is important to understand the processes of language formulation before the moment of language production. van Zaalen (2009) utilized Levelt's (1989) model of language production to explain the underlying processes and symptoms of cluttering. According to Levelt, the expression of ideas is a three-step process. The first step after the communicative intention deals with planning the idea or message and monitoring whether this is an accurate moment to express this message. The second step is the formulation of the message in correct grammatical sentence structures. The sentences are built with words that are gathered from the lexicon. Every word within the sentence has to be built up itself as well. Words are built with syllables. Syllables have to be pronounced in the right order (hence, "bi-bli-o-gra-phy" and not "bli-bi-gra-phy-o") and in the right way (not "bli-bli-o-gra-phy"). When the sentences and words are planned and a motor plan is ready, people can proceed to the third step by expressing their thoughts. A person with cluttering often speaks at a speech rate (articulatory rate plus pauses) that is not adjusted to linguistic or motor demands, resulting in a high frequency of normal disfluencies (a disruption of grammatical encoding), errors in pausing, and/or unintelligible words (a disruption of phonological encoding; St. Louis & Schulte, 2011; van Zaalen, 2009, 2022).

It is believed that PWC speak quickly, not allowing enough time for these formulation processes to effectively occur. In cluttering, the time in which all three steps of language production have to be finished is assumed to be under pressure, compared with the normal time frame (as indicated in Figure 1), and PWC are therefore probably

Figure 1. Mean articulatory rate (MeanArtrate) and mean pause duration (MeanPause) on three moments in time (first test session [T1], second test session [T2], and third test session [T3]). The green lines indicate the frame of preferred pause duration between phrases with lower (0.5 s) and upper (1.0 s) borders. The purple line indicates the mean articulatory rate for children before (4.4 syllables per second [SPS]) and during (5.6 SPS) adolescence.



prone to experience errors on the level of grammatical and/or phonological encoding. Condensed time can result in a high frequency of normal disfluencies, errors in pausing, and/or unintelligible words (van Zaalen, 2009).

At the same time, in our experience, we observed that when PWC focus on speech production and add longer pauses between phrases, they can be fluent and intelligible (van Zaalen, 2022; Winkelman, 1990). Most PWC are fluent and intelligible when reading aloud, likely because the writer already formulated the language.

Cognitive Component of Cluttering

On the basis of the biopsychosocial model, Stourneras (1980) described four components of stuttering: cognitive, emotional, verbal–motor, and communicative. These were adapted to cluttering by van Zaalen and Winkelman (2009). Under this adaptation, the cognitive component in cluttering is related to attention and habituation, specifically the attention capacity that PWC can use to monitor their speech and the thoughts people have about their (changed) communication (van Zaalen, 2007a).

The self-image of most people with fluent speech is generally moderately positive. Although some PWC are not aware that they clutter, they can be aware of negative reactions of others even before diagnosis. Therefore,

negative perceptions can develop just by listener reactions even in the absence of self-awareness of the PWC (Giuffre et al., 2021).

The increase in the speech rate of adolescents cooccurs with another important development: a growing level of self-reflection, especially regarding one's appearance and speech. When at first PWC are unaware of their disfluencies or unintelligibility, in adolescence, a vulnerable period in their lives, the gradual realization of speaking differently becomes more apparent. Often, this growing awareness develops because of feedback from people in their environment. Adolescents hear comments such as "Man, it's so hard to understand what you're saying"; "You better talk clearer"; and "What did you say?" (van Zaalen & Reichel, 2015).

On the basis of low symptom awareness, PWC generally judge their speech positively and are under the impression that listeners who have problems understanding them might not be paying enough attention to their speech. The original positive self-image changes for the worse as soon as the connection is made between listeners' negative feedback and the speakers' speech. Self-image mostly develops from the time of adolescence. Self-image is mostly disturbed in preadolescence in many areas: heightened self-consciousness; instability of self-image; slightly lower self-esteem; lower opinions of themselves

with regard to the qualities they valued; and reduced conviction that their parents, teachers, and peers of the same sex held favorable opinions of them (Simmons et al., 1973). Within families or schools, often-unsaid remarks exist. Expressions such as "What did you say?" or "He talks like a greyhound" are examples of those remarks. Although these remarks seem harmless, they can negatively affect a person's self-image, and we assume that they could have negative consequences in the end at a cognitive and social level (van Zaalen, 2007a).

Case Study: Johan

The Case of Johan

When Johan first entered our online clinic (due to COVID-19), our first impression was that of a vulnerable, shy young boy. He was not really interested in talking, only politely answering the therapists' questions with oneor two-word responses. His mum asked us to do an assessment because she thought he could be considered a person with cluttering. This diagnosis did not fit the boy we saw during my first sessions with Johan. However, this diagnosis was consistent with the boy we saw on at-home videos, talking with his father and siblings. Mum did her research online and named some of the key characteristics of cluttering reported in the literature: a fast and irregular rate, a high frequency of normal disfluencies and unfinished sentences in incomplete stories, loss of eye contact, and the appearance of decreased interest in communicating as noted in cluttered speech when language formulation takes too much attention capacity (van Zaalen, 2009). In addition, she noted the urge to talk about the same topic time after time. Because parents are an important source of knowledge, we trusted mum's gut feeling and decided to ask her to make recordings of Johan's speech in telling a story, reading aloud, and retelling the same story, as well as talking about a fun adventure. Mum sent us the recordings, and the analysis began. The primary conclusion we reached was that Johan did not adjust his speech to the language complexity of the moment, resulting in a high frequency of normal disfluencies, moments of unintelligibility, and a fast rate of speech (see Table 1). His lack of congruence between language and rate of speech was a clear indication of cluttering.

A careful assessment of the impact of the speech disturbances in the upcoming sessions led to the conclusion that Johan was not at all aware of any disturbance in his speech, but as he told us several times, "People do not

Table 1. Distribution of percentages	of fluent	and	disfluent	words	across	three	different	moments i	n
time, displayed per speech context.									

Percentage of words	NDFs	SLDs	aDFs	Fluent	
T1					
Reading	17.12 (SD = 5.5)	1.9 (SD = 1.7)	1.24 (SD = 1.4)	79.72 (SD = 7.6)	
Retelling	12.0 (SD = 0)	/		88 (SD = 0)	
Spontaneous	23.0 (SD = 0)	3 (SD = 0)	$ \begin{array}{c} 1\\ (SD = 0) \end{array} $	73 $(SD = 0)$	
М	17.12 (SD = 5.53)	1.9 (SD = 1.7)	1.2 (SD = 1.4)	79.0 (SD = 7.6)	
T2	(02 0.00)	(02)	(02)	(02)	
Reading	12.96 (SD = 3.7)	2.58 (SD = 1.90)	2.75 (SD = 0.4)	81.7 (SD = 1.9)	
Retelling	14.68 (SD = 3.1)	1.49 (SD = 0.4)	2.54 (SD = 0.2)	81.28 (SD = 2.5)	
Spontaneous	9.52 (SD = 0)	4.76 (SD = 0)	3.17 (SD = 0)	82.5 (SD = 0)	
М	`12.96 ´	2.58	2.75	` 81.7 ´	
Т3	(SD = 3.7)	(SD = 3.7)	(SD = 0.4)	(SD = 1.9)	
Reading	10.54 (SD = 6.65)	0.5 (SD = 0.6)	2(SD = 2.2)	86.96 (SD = 6.8)	
Retelling	` 5	` 1 ´	5	` 89 ´	
Spontaneous	(SD = 0.9) 10.17 (SD = 0)	(SD = 0.1) —	(SD = 0.2)	(SD = 1.1) 89.93 (SD = 0)	
М	10.54 (SD = 6.65)	0.5 (SD = 0.58)	2(SD = 2.2)	88.96 (SD = 6.8)	

Note. Em dashes indicate data not available. NDFs = normal disfluencies; SLDs = stutter-like disfluencies; aDFs = atypical disfluencies; T1 = first test session; T2 = second test session; T3 = third test session.

understand me or bother about my messages." Johan was "rather bored" by other people's stories. We hypothesized that this boredom was most likely due to his high total intelligence quotient, determined to be at 135 at the age of 11;7 (years;months). Because of his unawareness of the disturbances, we decided to start with indirect cluttering treatment without direct feedback on his speech output. Sessions aimed for complete 100-word stories about all types of topics (within and outside his interest area) and content monitoring. Feedback to Johan was limited to the number of used words in the story, the existence of pauses between sentences, story content, and the presence of eye contact.

Parent counseling at that time was supported by bibliotherapy, explaining the concept, the underlying mechanisms, and the prognosis of cluttering. Parents were mentally prepared for a significant increase in cluttering symptoms around the early start of preadolescence (11–13 years of age). Because of the expected natural increase in rate in the preadolescence phase and the unawareness of symptoms by Johan, not too much attention was placed on speech output in the early phases of therapy.

Johan started therapy at the age of 10 years. His articulatory rate was 5.07 SPS. Measuring the articulatory rate is a standard component of the Fluency Assessment Battery (van Zaalen & Reichel, 2014) and was regularly repeated during treatment sessions, as is part of cluttering treatment described by van Zaalen and Winkelman (2014) and van Zaalen (2022). When Johan was at the age of 10;8, his speech changed dramatically over a 2-week period. His articulatory rate in spontaneous speech, retelling, and reading increased drastically, resulting in an increase in normal disfluencies, short prolongations, and more than three moments per minute of coalescence.

Where Johan was not aware of any speech disruptions before, at the age of 10;8 (second test session [T2]), he most definitely was. He said, "My speech gets stuck" and "I do not want to talk anymore." After these remarks, he started to cry intensely but did not speak. Multiple moments of stutter-like disfluencies (SLDs) appeared, including tensed part-word repetitions and blocks with secondary behaviors such as eye blinking, pitch rise, and involuntary jaw movements. Johan indicated that he was not enjoying talking anymore. Although he previously spoke without hesitation, he now conversed in short, incomplete sentences only. When Johan became negatively aware of his disfluencies, his therapy addressed speech output directly. Audiovisual feedback (AVF) training (van Zaalen, 2007b; van Zaalen & Reichel, 2019) was used for different levels of language complexity. Although Johan was considered a fluent speaker before the age of 10 years, when he entered preadolescence, he became very disfluent and showed signs of avoidance. We will further examine what is known about the prevalence and onset of cluttering and its coexistence with stuttering.

Speech and Language Testing Protocol

The first author assessed Johan's speech and language at three time points (first test session [T1], second test session [T2], and third test session [T3]). Assessment of Johan was done at the ages of 10;0 (before treatment of storytelling skills), 10;8 (at the start of AVF training), and 11;2 (during AVF training). T1 took place when Johan was 10 years old, 1 month after Johan's mother discovered the existence of cluttering online. T2 occurred when he was at the age of 10;8, 1 week after Johan started stuttering and avoided speaking. T3 took place 3 months later at the age of 11;2. Between T1 and T2, twelve 45min online training sessions focused on storytelling skills. Between T2 and T3, AVF training (van Zaalen & Reichel, 2019; van Zaalen & Winkelman, 2014) was conducted, with 45-min online training sessions twice a week and everyday home monitoring tasks.

All speech and language examinations were videorecorded. Speech samples were collected during spontaneous speech as well as reading, describing, telling, and retelling "The Wallet Story" (van Zaalen & Bochane, 2007). For each test session, at least 350 words of Johan's spontaneous speech were transcribed for off-line fluency and rate analysis using Praat speech analysis software (Boersma & Weenink, 2021).

Disfluency analysis was done by differentiating normal, stutter-like, and atypical disfluencies. The disfluencies were measured as percentage of spoken words and were rated by both authors in order to assess interrater reliability using the "percent agreement index" (Cordes et al., 1992). Reliability measures on the total disfluency frequency were 98% for telling, retelling, and reading. The normal disfluencies explored in this study are those indicated by Myers et al. (2012). These normal disfluencies are part-word, whole-word, and phrase repetitions; interjections; and revisions. Prolongations; blocks; and sound, syllable, and tensed monosyllabic-word repetitions were identified as moments of stuttering. Atypical disfluencies explored in this study are those that do not fit in the categories of normal disfluencies or SLDs. These disfluencies included mid- or end-word repetitions. Atypical disfluencies have been observed in different conditions, such as in people with intellectual challenges, autism spectrum disorders, neurofibromatosis type 1, acquired neurological damage, and genetic syndromes (Coppens-Hofman et al., 2013; Cosyns et al., 2010; Scaler Scott et al., 2014; Sisskin & Wasilus, 2014). Definitions of these kinds of disfluencies slightly vary from author to author. In Johan's speech, they were defined as mid-word repetitions of sound or syllable and end-word repetitions of sound or syllable, both without visible/audible tension. Apart from normal, stutter-like, and atypical disfluencies, Johan also exhibited telescoping or coalescence of words, which is a cluttering symptom related to speech rate (van Zaalen & Reichel, 2015).

Rate and pause measurements were done using Praat speech analysis software based on the rate measurement protocol described by Cosyns et al. (2014) and van Zaalen (2010). In addition, Johan completed the Speech Situation Checklist–Emotional Reaction (SSC-ER; Brutten & Vanryckeghem, 2003). Finally, the MAR and mean pause duration between phrases were measured, as described by van Zaalen-op't Hof et al. (2009a), Hall et al. (1999), and Cosyns et al. (2018).

Therapy Approach for Johan

PWC are often unaware of the speech disruptions as they occur. The act that other people mention that PWC talk too fast, are disfluent, or are unintelligible is often not internalized in a manner necessary for treatment. AVF training has proved to be effective on both the short and the long term for treating cluttering (van Zaalen & Reichel, 2019). In AVF training (see Supplemental Material S1), recordings of PWC are played back while being displayed via Praat speech analysis software on a computer screen. In listening to and looking at their speech over and over again, combined with an analysis of the rate, fluency, pause placement, and duration, the client observes and hears what a listener sees and hears. Instead of arguing about the rate being too fast or the speech being disfluent, in AVF training, speech is measured using Praat software. Such an approach is based not on subjective opinion but on objective analysis. Each therapy session of 1 hr is done according to a strict protocol, as follows: (1) The goal of the recording exercises is determined. (2) Spontaneous speech is recorded using a digital audio recording and the speech analysis software Praat (Boersma & Weenink, 2021). (3) While recording, the time of significant moments is written on a piece of paper by the clinician. (4) The recorded file is opened using Praat speech analysis software and played and displayed. (5) PWC are given narrow observer instructions to specific aspects of speech production (e.g., "Are you able to hear all the syllables in this piece?"). (6) A significant moment is played back (i.e., a segment of a maximum of 20 s), in which the goal set for the client was not reached. (7) Characteristics such as articulatory rate or pause duration are measured, and results are discussed. (8) Steps 6 and 7 are repeated with two fragments that did not go well and three fragments that did go well. (9) The exercise is repeated, and results of the first recording are compared with results of the second recording. (10) Home assignments are formulated. At home, the PWC make five recordings each day and analyze two of these recordings by themselves. Analyses of the home recordings are discussed in the next session. After the PWC become aware of their speech disruptions, AVF training focuses not on moments of disfluency or reduced intelligibility but on moments of adequate pausing or on a normal articulatory rate. The clients learn what they can do to change their speech and how it sounds,

looks, and feels when their speech is accurate, fluent, and intelligible.

For the AVF training, positive feedback is essential. For example, the clinician may write down the successful segments of speech while recording the speech. When playing back these pieces with the client, the clinician can then say something such as the following: "Listen, this is very intelligible and when we look at it, we can clearly see all syllables on the screen, you can hear every syllable of the word, and it also sounds natural"; "Let us listen again because this is very good"; "Yes, this is what we were hoping for"; or "Let us measure your (articulatory) rate so we know at which rate you can be really intelligible." The clinicians' focus on moments of success helps clients build stronger speech control and increases the clients' confidence and future successes.

The AVF training (van Zaalen, 2022) makes it possible to correctly respond to the speech characteristics and to create a frame of reference in which the speech is intelligible and fluent (van Zaalen & Reichel, 2015). AVF training has been found to give positive results on cognitive, social–emotional, and speech motor characteristics (van Zaalen & Reichel, 2019).

Results

SLDs

No early onset of stuttering emerged in Johan's history. At T1, Johan did produce a mean of 1.9% (SD=1.7) SLDs in the speech tasks. At T2, Johan produced a mean of 2.58% (SD=3.7) SLDs. Although Johan showed some SLDs at T2, the frequency and the duration of these SLDs were low overall and compared with his normal disfluencies. Johan's stuttering frequency showed a higher relative frequency of tensed part-word repetitions in spontaneous speech at T2 compared with that at T1.

At T3, Johan produced a mean of 0.5% (SD = 0.58) SLDs, with a very short duration (< 0.3 s). Prolongations and blocks were very rare, and if they occurred, the duration was < 0.3 s. Please see Table 1 for results.

Frequency of Normal Disfluencies

At T1, Johan produced normal disfluencies on 23% of the words in spontaneous speech, on 18% when retelling a story, and on 13% while reading, resulting in a mean of 17.1% across speech tasks. At T2, Johan produced normal disfluencies on 9.52% of the words in spontaneous speech and on 14.68% during reading, resulting in a mean of 12.96% across speech tasks. At T3, Johan produced normal disfluencies on 10.17% of the words in spontaneous speech, on 5% when retelling a story, and on 10.54% during reading, resulting in a mean of 10.54% across speech tasks. Please refer to Table 1 for results.

Atypical Disfluencies and Cluttering Symptoms

At T1, T2, and T3, Johan showed both mid- and end-word repetitions in his spontaneous speech. At T1, in reading a text, mid- and end-word repetitions were observed in words that were multisyllabic, difficult to pronounce, and new to him, such as "Massachusetts." At T1, the mean of atypical disfluencies was 1.2%; at T2, it increased to 2.75% and then lowered to 2% at T3 (see Table 1). The types of atypical disfluencies did not change over time. In retelling a story, some moments of coalescence/telescoping (T1 = 2, T2 = 0, T3 = 0) and self-corrections (T1 = 0, T2 = 0, T3 = 2) were noticed.

Disfluency Types and Speech Context

The mean of fluent words at T1 was 79% (SD = 7.6) and increased to 81.7% (SD = 1.9) at T2 and 88.96% (SD = 6.8) at T3. During spontaneous speech, 81% (T1), 78% (T2), and 91.5% (T3) of Johan's disfluencies were normal disfluencies, specifically repetitions of syllables, words, or phrases. Most typically, words or phrases were repeated once, but occasionally, the repetitions could be up to four fast word repetitions in the rhythm of speech (e.g., "I.I.I. really want to tell about it") with, at T2, twice increased tension and one pitch rise (in this case, an emotional response unrelated to stuttering). Part-word repetitions (tensed), prolongations, and blocks were present in a minority of the disfluent moments. The distribution of the disfluency types during different levels of language complexity was comparable over time. Please see Table 2 for results.

We observed most normal disfluencies such as word part repetitions, phrase repetitions, revisions, and interjections in the medial position of the sentence, whereas word repetitions were observed in both the initial and medial positions of the sentence. No detailed analyses of this were done within the scope of this project. In our clinical experience, this is typical for cluttering and contrary to the disfluencies in stuttering that normally happen at the beginning of the sentence, for example, "I went to the, I walked to the shop" or "Just because I I really want to, to, to think about the final event of today." To our knowledge, no disfluency studies exist indicating and understanding

the position of cluttering disfluencies in the sentences. The remaining disfluencies were atypical disfluencies presented at T1 and T2 as within- or end-word syllable repetitions (see Table 2).

Secondary Behaviors

At T1, eye blinking was noticed once during a tensed part-word repetition. At T2, Johan used prolongations with a pitch rise during mazing, on two occasions when he was stuck on words that were very difficult to read (e.g., "Macca...Massa...Maccashushs" when he wanted to read "Massachusetts"). At T3, no secondary behaviors were observed.

Mean Pause Duration and MAR

The MAR increased from 5.07 SPS at T1 to 5.83 SPS at T2 and further increased to 6.08 SPS at T3. At all times, Johan's articulatory rate was faster compared with those of his peers (Amir & Grinfeld, 2011; van Zaalen, 2022). At the same time, the mean pause duration remained too short at T1 (0.019 s) and T2 (0.032 s), whereas at T3, it was on an adequate level (between 0.5 and 1.0 s; van Zaalen, 2009, 2022). Please see Figure 1 for results.

Emotions and Attitudes

At T1, Johan was not aware of any disfluencies. He showed a positive attitude toward communication, and he did not avoid speaking. His scores on the SSC-ER (Brutten & Vanryckeghem, 2003) were comparable to those of nonstuttering subjects, and his scores fell into the category of "Unaware/Unconcerned." The mean of fluent words at T1 was 79.5%.

At T2, Johan was aware of his disfluencies and avoided speaking situations. He spoke in short sentences with sporadic blocks and prolongations with tension. The mean of fluent words at T2 was 70.3%. His scores placed him in the category of "Aware/Concerned."

At T3, Johan was aware of his disfluencies and was able to control his speech whenever he was bothered by too many disfluencies. This was noted on the first four levels of language complexity (naming, reading, telling,

Table 2. Type and relative frequency of disfluencies per time slot in spontaneous speech.

Test session		Type and relative frequency of disfluencies									
	Normal					Stutter-like			Atypical		
	WPR	WR	PR	REV	INT	pWR	PRO	В	MwR	Endwr	
T1	7.4	18.5	18.5	14.8	25.9	7.4	_	3.7	3.7	_	
T2 T3	0 50.0	18.2 30.0	9.0 16.7	17.8 —	9.1 —	27.3 —	_	_	_	9.1 —	

Note. Em dashes indicate data not present/zero. WPR = word part repetitions (without tension); WR = word repetitions; PR = phrase repetitions; REV = revisions; INT = interjections; pWR = tensed part-word repetition; PRO = prolongations; B = blocks; MwR = mid-word repetitions; Endwr = end-word repetitions; T1 = first test session; T2 = second test session; T3 = third test session.

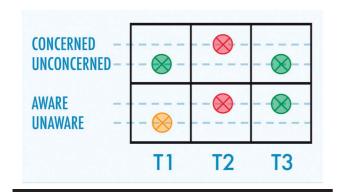
and retelling). Because he understood what happened in his communication, now being able to control his rate and cope with the disfluencies, his scores placed him in the category of "Aware/Unconcerned." The mean of fluent words at T3 was 77.8%. Please see Figure 2 for results.

Discussion and Conclusions

Cluttering Severity

The frequency of normal disfluencies in fluent speakers is reported to range between 3.1% (De Nil et al., 2005) and about 9.7% (Blokker et al., 2010; Eggers et al., 2010) in a group of young children and adolescents. In some PWC, normal disfluencies can be as frequent as 35% (van Zaalen-op't Hof et al., 2009c). In contrast, people who stutter tend to produce fewer than 5% normal disfluencies (van Zaalen-op't Hof et al., 2009c). Howell and Au-Yeung (2002a) suggest that planning and execution happen in parallel. Planning of the linguistic formulator can be under time pressure when a segment that is difficult and, therefore, time consuming to generate has to be prepared, and this plan is required quickly as when the planned segment follows a word that is executed rapidly (Howell & Au-Yeung, 2002a, p. 78). In a fast rate of speech, as known for cluttering, it is possible that the linguistic formulator can fail to deliver the complete plan on time, resulting in normal disfluencies. More research on self-corrections in cluttering is needed to confirm this suggestion of dissynchronicity between planning and execution in cluttering. Johan produced a high frequency of normal disfluencies at T1 and T2. Therefore, his cluttering can be diagnosed as mainly syntactic cluttering (see the introduction; van Zaalen & Winkelman, 2014). Although Johan experienced more speech control at T3, the amount of normal disfluencies was still higher compared with that of fluent controls. We assume that the combination of

Figure 2. Visualization of awareness of disfluencies and communicative concern in three moments in time (first test session [T1], second test session [T2], and third test session [T3]).



longer pauses between phrases, higher monitoring skills, and a lower frequency of disfluencies gave him a sense of speech control again. This illustrates that cluttering severity cannot be based on a single variable in speech production.

Johan's inhibition decreased from T1 to T2, as indicated by the diminishing duration of pauses between sentences, bodily restlessness during speech, and his ongoing willingness to debate and discuss. Given the changes to executive functioning previously mentioned in adolescence, inhibition of behavior may be viewed as a physiological subcomponent of cluttering during preadolescence and adolescence. Stopping an ongoing activity and switching to a new one if necessary is difficult. When speech is initiated, it is hard for PWC to stop it. Although the verbiage of people who clutter demands many of the listener's skills, the lack of inhibitory control does not disturb or stop the person who clutters. At T2, the pauses between sentences were almost diminished, whereas at T3, they were comparable to those at T1. At T2, Johan was not only speaking faster, but his pauses between sentences also diminished, leading to an increase of both normal disfluencies and SLDs as well as lack of control, which frustrated him. The lack of inhibition in PWC is also noticeable in their bodily restlessness during speech, but not during listening (van Zaalen & Reichel, 2015). This restlessness in communication can distract the listener from the message as well. When the pauses between sentences are needed to process the speech of the speaker and a speaker lacks accurate pauses, the listener's perception is impeded.

Normally, a person detects errors in the pauses between words or sentences and then repairs speech errors. We concluded that although Johan was able to recognize speech errors in his recorded speech at T3, at T1 and T2, he was unaware of such errors in running speech. In running speech, Johan was unable to pay enough attention to such errors. After direct intervention with AVF, Johan became aware of the errors and was able to repair them. AVF training helped Johan in two ways. On the one hand, the training helped him better monitor his speech. On the other hand, AVF training helped Johan understand the variability of his speech performance and realize that although the story in his mind is clear, he has to monitor his speech for the listener to understand him.

Cluttering Awareness

The fact that Johan became aware of his speech disfluency may be allocated to his listeners' responses (e.g., imitation of disfluencies or ignoring his statements) and to the decrease in overall fluency. Another explanation of Johan's awareness of cluttering is that between T1 and T2, his cluttering severity became higher. Cluttering severity is influenced by qualitative variations (revisions vs. word repetitions, appropriate vs. inappropriate loci of pauses; Bakker & Myers, 2014)

and the degree of telescoping or coalescence (van Zaalen, 2009; Ward, 2018). All of these are strongly influenced by the levels of language complexity, rate modulation, and articulatory precision. Cluttering is therefore seen as a perceptually based phenomenon, that is, "a package deal" not easily amenable to frequency counts (Bakker & Myers, 2014).

Adolescents with cluttering understand that they are doing something wrong but are not aware of exactly what it is that they are doing wrong. Therefore, they cannot change their speech behavior and tend to become insecure in their speech performance. PWC who are prone to be cluttererstutterers can show SLDs (St. Louis et al., 2007; Ward, 2006, 2018; Winkelman, 1990). We argue that the problems in inhibition influence speech motor control. We explain this with the saying: "You stumble (clutter) before you fall (stutter)." When the rate of speech is too fast, PWC tend to tumble over their words but continue to talk fast. In some cases, if the person is also prone to stutter, it is possible that the motor system is overloaded with a block or prolongation because of that. Because these disfluencies appeared sporadically in Johan's case, we can consider Johan as being a clutterer-stutterer who is mostly presented as a clutterer.

Until now, little research has been conducted in the adolescent age group of both fluent and disfluent populations. To understand the changes in preadolescence, longitudinal research is needed in which disfluent and fluent children are followed before preadolescence, during adolescence, and after adolescence. Eventually, in order to avoid the social–cognitive–emotional consequences as described in Johan's case, early treatment is recommended as soon as a negative communication attitude arises.

Treatment

On the basis of his response at T2, we believe that if Johan had not received the appropriate therapy, his speech would have been prone to being perceived as extremely fast and unintelligible to others, and he would lose the ability to get his message across, with severe negative consequences to his self-esteem. Given Johan's case, this is especially important for speech-language clinicians to be aware of when clients are approaching preadolescence and adolescence. AVF training is an example of one approach to enhance speech control and improve speech monitoring (van Zaalen & Reichel, 2019). Apart from these speech-related goals, speech-language therapists should focus on increasing the clients' understanding of cluttering and social participation and on putting needed accommodations in place to stabilize school performance.

Limitations

In order to be sure that Johan's natural speech was examined without him adding more focus to his speech,

the recordings of spontaneous speech analyzed were not made in a standardized context. The symptoms that occur in cluttering increase with time pressure and relaxation and decrease with focus. Recordings in a fixed environment would have given an unrealistic image of his performance. We do understand that a comparison between different moments is a limitation of this choice; therefore, we analyzed and compared conversations within levels of language complexity and used the same reading book in all recordings. Two independent researchers analyzed the recordings individually using Praat speech analysis software (Praat.org) to ensure an objective analysis of speech. Praat software allowed for the identification of all produced syllables and barely audible word repetitions.

Measuring the MAR was done on 10–20 fluent consecutive syllables without pauses. Determining the articulatory rate can be highly affected in speakers who experience many disfluencies; they simply do not produce enough consecutive fluent syllables without pauses. Johan produced spurts at a rate that was sometimes so fast he was barely intelligible. Therefore, the fastest productions could not be used in the analysis. Measuring the speech rate instead of the articulatory rate would not have solved this issue but would add to the difficulty of dealing with all the extra pauses in linguistically incorrect places. Further research on this and on the relation between pauses, rate, and disfluencies is needed to better understand their correlation.

Conclusions

AVF training helped Johan when he became negatively aware of his disfluencies. In preadolescence, cluttering can drastically change a young child's life. While being unaware of their speech condition before adolescence, during preadolescence, the changes in brain organization lead to an increase in rate and a decrease in speech control. Not understanding what is happening, a negative communication attitude is at risk. Speech-language therapists are strongly advised to monitor children with cluttering signs in the early years of their adolescence.

Data Availability Statement

Data supporting the results are available and can be reused. Data can be accessed by sending an e-mail to the corresponding author, and she will send the data file within 1 week.

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and, thus, give very important insights to the scientific community and other parents about cluttering in adolescence.

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