Research Article

Evidence for Stuttering Disfluencies to be Both the Result of Impaired Speech Production System and Coping Response to the Impairment

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Abstract

The present study analyzed the topography of recurrent stutters (stutters on same words in repeated readings) in a group of 22 adolescents and adults who stutter to explore whether stutters are the direct result of impaired speech production system or coping responses to an underlying impairment. If stutters are the direct result of an impairment, there should be more concordant recurrences (stutter of a certain type on a word in one reading is followed by stutter of the same type if the word is stuttered in the next reading of the passage) than discordant recurrences. Significantly greater number of discordant recurrences suggests that stutters may be coping responses to an underlying impairment. The present study found significantly more concordant than discordant recurrences indicating at least some stutters are the direct result of impaired speech production system. Results of the study also suggest that non-repetitive stutters (dysrhythmic phonations) may be coping responses.

Introduction

Stutter Topography

Topography means the features and configuration of a geographic area or a physical or behavioral entity. Topography, in the context of stuttering, refers to the observable (audible and visual) features and configuration of a relatively discrete instance of stuttering, which is sometimes referred to as the "moment of stuttering" (Bloodstein & Bernstein Ratner, 2008). Johnson (1959) listed eight disfluencies that might be observed in both people who stutter (PWS) and those who do not (PWNS). The list included interjections, part-word repetitions, word repetitions, phrase repetitions, revisions, incomplete phrases, broken words, and prolonged sounds. Wingate (1964) contended that audible or silent repetition of sounds, syllables, and one-syllable words and audible and silent prolongation of sounds are sufficient to define the speech characteristics of stuttering. Silent repetitions and prolongations refer to repetitive and static articulatory postures respectively with no audible sound. Yairi and Ambrose (1999) described part-word repetitions (PWR; repeated production of a portion of a word), single-syllable word repetitions, and dysrhythmic phonations (DP) as "stuttering-like disfluencies" or SLDs. DP"....includes sound prolongations, silent blocks and also broken words and other within-word interruptions (but not repetitions) that disturb the continuity of words" (Yairi & Ambrose, 1999, p. 1100). Other types of disfluencies on Johnson's list were regarded as not typical of stuttering.

Wingate, Yairi and Ambrose appear to be in general agreement as to the topography of a moment of stuttering. Teesson, Packman, and Onslow (2003) proposed a scheme for describing stutters, which they called the Lidcombe Behavioral Data Language (LBDL). Speech characteristics included as part of stutters in LBDL are repeated movements (syllable repetitions, incomplete syllable repetitions, and multisyllable unit repetitions) and fixed articulatory postures (with and without audible airflow). Repeated movements category in LBDL comprises all repetitions included in the Wingate and Yairi and Ambrose taxonomies and, additionally, also includes multisyllable word repetitions and repetition of phrases and entire utterances. This is at variance with what is generally regarded as stutters by most researchers (see Tables 12.3a and 12.3b, p. 315 and associated

discussion in Bloodstein & Bernstein Ratner, 2008; Young, 1984). Overall, however, there appears to be more agreement than disagreement as to what constitutes a stutter.

A causal theory of stuttering must necessarily account for its topography (Packman & Attanasio, 2004). A theory or model of stuttering must explain why stutters occur and why they take the form they do. Kolk (1991) pointed out that theories of stuttering fall into two broad types based on how they explain stutter topography. Certain theories hold that stutters are the direct result of an impairment. For instance, the neuropsycholinguistic theory of stuttering (Perkins, Kent & Curlee, 1991) states that stutters occur when the rate of speech "....exceeds the rate at which segments can be integrated synchronously into their syllable frames" (p.748). Other theories attribute stutters to interhemispheric interference (Forster & Webster, 2001), or to ".... an inadequacy of neuronal resources for sensory-motor information processing" (Neilson & Neilson, 1987, p. 325) or some other motor, sensory, linguistic, or psychological factor, singly or in some combination (e.g., Karniol 1995; Packman, Onslow & Menzies, 2000; Venkatagiri, 2005).

Certain other theories maintain that stutters are coping (adpative) responses and not the direct consequence of an impairment. Stutters are viewed in these theories as representing a conscious or unconscious (and often futile) attempt by the speech production system to overcome or circumvent an impairment that interferes with fluent speech production. It is almost universally accepted that most of the older children and adults who stutter are constantly trying to minimize or otherwise control the occurrence or severity of stutters when they are talking. These attempts involving movement of body parts far removed from the speech apparatus, extraneous sounds unrelated to speech being produced, etc. are variously referred to as "secondary behaviors" (Van Riper, 1982) or "accessory features" (Wingate, 1964), or "superfluous behaviors" (Teesson, Packman & Onslow, 2003). However, three relatively influential theories of stuttering explain the core features of stuttering - sound and syllable repetitions, sound prolongations, and articulatory postures - as resulting from an attempt to mitigate an underlying psychological or neurolinguistic impairment.

Bloodstein (1997; 2001) has offered the anticipatory struggle hypothesis of stuttering, which, in its most recent formulation (Bloodstein, 2001), holds that in developed stuttering (as opposed to incipient stuttering), stutters are the product of haphazard and tension-filled effort to overcome or circumvent an anticipated difficulty in speaking based on a false belief that speaking is difficult and failure is imminent. Kolk and Postma (1997) have presented the covert repair hypothesis, which states that stutters are a "....'normal' repair reaction to an abnormal phonetic plan" (p. 193). According to this theory (or hypothesis), the utterance plans prepared by the PWS are replete with errors and stutters result when the PWS attempt to repair them covertly (before the utterances are spoken). Like Bloodstein (2001), Howell (2004) also distinguishes between beginning stuttering in children and developed stuttering in adults in his EXPLAN theory of stuttering. EXPLAN theory hypothesizes that PWS are slow to prepare the linguistic and motor plans for the longer and more complex content words (nouns, verbs, adjectives, and adverbs). Children in the beginning stage of stuttering repeat shorter and simpler function words (articles, prepositions, conjunctions, and pronouns) that precede content words to gain more time for preparing the plans for the content words. Therefore, in the beginning stage of stuttering, stutters are adaptive responses to compensate for the slower linguistic and articulatory planning of certain words. In developed stuttering in older children and adults, the speakers do not repeat function words but try to execute the as yet linguistically or articulatorily incomplete content words. This premature execution produces stutters, which are directly caused by the impaired (slower than normal) speech planning system.

Whether stutters are the result of an impairment or a coping response to an impairment is a fundamental question that must be answered in order to advance our understanding of stuttering. Nearly all experimental research studies employ a description or count of stutters as a dependent variable and attempt to relate them to the experimental manipulation of independent variable(s). The same findings may be interpreted in two different ways depending on how one views stutters. However, few studies have directly addressed this conundrum and, according to Packman and Attanasio (2004), many theories completely fail to address the topography of stuttering while others provide explanations that lack empirical and rational bases. The present study is a preliminary attempt to address this admittedly difficult problem.

Recurrent Stutters

When PWS read a passage repeatedly two or more times, a certain number of words are likely to be stuttered more than once in successive readings (Johnson & Knott, 1937; Johnson & Inness, 1939; Wingate, 1986). This observation - the same words being stuttered more than once in repeated readings - was referred to as the consistency of stuttering (Johnson & Knott, 1937). Wingate (1986) objected to the term "consistency" to refer to this phenomenon on the grounds that loci of stutters in repeated readings do not show the high degree of reliability, stability, and regularity denoted by the term. Instead he preferred to call this phenomenon "recurrence." "Consistency" is an evaluative and explanatory term whereas "recurrence" is a descriptive term. We will adopt "recurrence" in this paper.

Stuttering theories tend to attribute the occurrence of different stutter types at different times to differences in linguistic (vocabulary, sentence structure, etc.), communicative (attentive or inattentive listener, audience size, etc.), psychological (level of anxiety, time pressure, etc.), and physical (supportive home environment versus difficult work environment, etc.) variables. However, if stutters are the direct result of a neurolinguistic or psychological impairment, there is a high probability that recurrent stutters would be concordant for type. If the stutter in Reading 1 was a PWR, then a stutter occurring on the same word in nearly identical physical, psychological, linguistic, and communicative context moments later in Reading 2, would also likely to be a PWR. If this were the case, then there should be a high degree of concordance for type among recurrent stutters. On the other hand, if stutters are coping responses to an underlying impairment, the recurrent stutters are more likely to be discordant for type. This is because coping responses tend to be variable as the system tries to find effective ways to minimize or get around the difficulty encountered during speech production.

The only substantive difference between two successive readings is the degree of linguistic and motoric familiarity with the reading material. There is a steady decrease in the frequency of stutters in repeated readings, known as the adaptation effect, although the stutter frequency plateaus after three to five readings (Johnson & Knott, 1937; for recent work on this phenomenon, see Bloodstein & Bernstein Ratner, 2008). Familiarity may reduce the frequency and severity of recurrent stutters (a three-unit syllable repetition may become a two-unit syllable repetition or a two-second sound prolongation may change to a one-second prolongation) but it is less probable that a PWR will turn into a DP or vice versa in the second attempt if the stutter is the direct result of an impaired speech production system. On the other hand, a high level of discordance for type in recurrent stutters suggests that the speaker consciously or unconsciously attempted to alter his/her speech because the preceding attempt to deal with an underlying impairment did not succeed.

There is, however, another possibility - some stutters may be the direct result of an impairment while other stutters may represent what the speaker does to cope with it. It is widely accepted that stutter topography generally changes over time (Bloodstein, 1960). Near the onset of stuttering, the most common type of stutter displayed by PWS is sound and syllable repetitions including words of one syllable (Meyers, 1986; Yairi & Lewis, 1984). Nonrepetitive stutters such as dysrhythmic phonations are less frequent in early stuttering. It is, therefore, possible, that repetitive stutters are the direct result of the underlying impairment whereas the nonrepetitive stutters represent the coping responses developed over a period of time. If this is true, repetitive stutters should show a high degree of concordance and nonrepetitive stutters should display a high degree of discordance.

Research Questions: For the purposes of this study, two broad types of stutters are distinguished - repetitive stutters (repetition of a sound, syllable, or multiple syllables of a word but not entire words or phrases) and non-repetitive stutters (dysrhythmic phonations including sound prolongations). The present study is designed to answer the following questions:

1. What is the mean percentage of concordance (i.e., stutters are of similar types) and discordance (stutters are of different types) in pair-wise comparisons (stutter on a word in Reading 1 is compared with stutter on the same word in Reading 2, stutter on a word in Reading 2 is compared with stutter on the same word in Reading 3 and so on) in five repeated readings of a prose passage? A statistically significant concordance is indicative of stutters, for the most part, being the direct result of an impaired speech production system whereas a statistically significant discordance suggests that they may be coping responses to an underlying impairment.

2. What is the mean percentage of concordance and discordance for repetitive and non-repetitive stutters separately in pair-wise comparisons in five repeated readings of a prose passage? A statistically significant concordance for either type of stutter points to its being the direct result of an impaired speech production system whereas a statistically significant discordance for either type of stutter is suggestive of its being an adaptive response to an underlying impairment.

Method

Participants

A group of 24 persons who stutter (PWS) participated in the study. Due to technical difficulties, the audio data recorded for one participant was unusable. In addition, a portion of the audio data for one of the participants for one of the four passages in Reading 1 could not be analyzed. The findings are based on the data for 23 participants, all males, ranging in age from 13 to 30 years with a mean age of 18 years (SD = 4.27). The criteria for inclusion in the study were as follows: (1) Native speaker of Kannada language and be able to read Kannada newspapers and magazines; (2) Diagnosed as a person who stutters by a qualified (Master's or Ph. D level) speech-language pathologist with clinical experience in stuttering; (3) No self- or parent-reported history of hearing impairment, neurological problems, or language delay; (4) Normal orofacial structures and functions. Based on case history, an inventory of associated features, and

an analysis of spontaneous conversational speech, the stuttering severity was classified as mild (7 participants), moderate (11 participants), and severe (5 participants). None of the participants had received treatment for their stuttering at the time of the collection of data. The study was approved through an institutional review process as meeting the ethical standards and informed consent was obtained from either the participant or his parent in the case of the minor.

Procedure

Each participant read each of the four short passages five times in succession. The four factual passages together contained 194 words and each participant read a total of 970 words (5 X 194). Their readings were audio-recorded using Sony ICD-P320 digital recorder set to HQ Recording Mode (19.2 Kbps). The participant was seated across the experimenter in a clinic room and given a printed passage to read. The microphone of the recording device was kept approximately 10 cm from the mouth of the participant. The instruction, given in Kannada, was as follows: "Please read this passage aloud as you normally would. Are you ready? Now start." There was no pause between successive readings of a passage and only a brief pause, sufficient to hand the next passage and repeat the instruction, was present between passages.

Data Analysis

The recorded data was transferred to a laptop computer (Compaq Presario tu770) for analysis. Using the Audacity software program (version 2.0.3; http://audacity.sourceforge.net/), stuttered words were marked on printed copies of the reading passages. Audacity has the ability to playback speech at a slower rate without altering pitch to aid in the accurate marking of stutters. Part-word (audible sound or syllable) repetitions (PWR) (Wingate, 1964) and dysrhythmic phonations (DP) were marked separately. DP included one or more of the following subtypes: audible sound prolongations; tense pauses (pauses between words); and broken words (a period of silence in the middle of a word) (Yairi & Ambrose, 1999). Stutters marked as having both a PWR and a DP (39 instances) were excluded from further analysis.

Reliability

The inter-judge reliability was computed by pseudo-randomly selecting a subset of passages read by each participant. On average, 300 words drawn from the five readings (roughly a third of the total number of words read by each participant), were independently analyzed by the first author. The inter-judge reliability was computed using the Agreement Index described by Sander (1961) separately for part-word repetitions and dysrhythmic phonations. Sander's formula is widely used to determine inter-judge reliability in stuttering literature. The mean inter-judge reliability for PWRs was 0.96 with a range of 0.89 to 1.00 for the 23 participants. The mean inter-judge reliability for DPs was 0.92 with a range of 0.82 to 1.00 for the 23 participants. The inter-judge reliability was also computed using the Kappa statistic (Cohen, 1960) to determine the word-by-word agreement for stutter count between the original analysis and the second analysis. The mean Kappa was 0.872 ($p < 0.001$) with a range of 0.76 to 0.94. For intra-investigator reliability, approximately 25% of audio recordings of readings from each of the 23 participants were reanalyzed by the person who originally identified the stutters (second author or an assistant who is also a qualified speech-language clinician with a Master's degree). The mean intra-investigator reliability, using Sander's formula, was high; for PWRs, 0.99 with a range of 0.94 to 1.00 and for DPs, 0.98 with a range of 0.91 to 100 for the 23 participants.

Results

Stutter and Recurrence Rates

The data from one participant was excluded from further analysis because he had a very low, 2.29 percent of words stuttered with no recurrent stutters in adjacent readings. Therefore, the following analysis is based on data for 22 participants. The mean rate of stutters for the group was 13.39 percent of 970 words read (SD = 10.86) with a range of 3.09 to 47.53 percent. The mean percent of repetitive (sound and syllable) stutters was 7.68 with a range of 1.55 to 33.09 percent (SD = 7.89). The mean percent of non-repetitive (dysrhythmic type) stutters was 5.71 with a range of 0.62 to 21.34 (SD = 4.53).

On average, there were 42.18 recurrences (stutters on adjacent pairs of readings) per participant with a range of 2 to 222 (SD = 55.17). Participants with high rates of stutters also had high rates of recurrences and vice versa. The mean number of R - R recurrences (a repetitive stutter was followed by a repetitive stutter) per participant was 15.73 with a range of 0 to 119 (SD = 27.25) and the mean number of R - NR stutters (a repetitive stutter was followed by a non-repetitive stutter) was 8.50 with a range of 0 to 45 (SD = 10.34). The mean number of NR - NR recurrences (a non-repetitive stutter was followed by a non-repetitive stutter) per participant was 10.14 with a range of 0 to 61 (SD = 13.69) and the mean number of NR - R stutters (a non-repetitive stutter was followed by a repetitive stutter) was 7.82 with a range of 0 to 57 (SD = 12.65). Because the rate of concordant (a stutter was followed by a stutter of the same type) and discordant (a stutter of a certain type was followed by a stutter of a different type) recurrences varied considerably both within and across participants, Table 1 presents concordant and discor-

Table 1: The percent of concordant and discordant recurrences computed against total recurrences (R = repetitive stutter; NR = non-repetitive stutter)

Participants	Total concordant recurrences	Total Discordant Recurrences	R-R (Concordant) Recurrences	R-NR (Discordant) Recurrences	NR-NR (Concordant) Recurrences	NR-R (Discordant) Recurrences
1	60.00	40.00	60.00	20.00	0.00	20.00
2	60.00	40.00	10.00	10.00	50.00	30.00
3	58.54	41.46	31.71	21.95	26.83	19.51
4	57.84	42.16	46.08	18.63	11.76	23.53
5	0.00	100.00	0.00	50.00	0.00	50.00
6	100.00	0.00	75.00	0.00	25.00	0.00
7	54.05	45.95	26.58	20.27	27.48	25.68
8	50.00	50.00	50.00	50.00	0.00	0.00
9	74.07	25.93	48.15	18.52	25.93	7.41
10	73.68	26.32	69.59	14.04	4.09	12.28
11	62.50	37.50	12.50	18.75	50.00	18.75
12	88.57	11.43	2.86	5.71	85.71	5.71
13	54.24	45.76	15.25	23.73	38.98	22.03
14	66.67	33.33	16.67	16.67	50.00	16.67
15	68.18	31.82	36.36	13.64	31.82	18.18
16	58.06	41.94	35.48	25.81	22.58	16.13
17	61.11	38.89	38.89	33.33	22.22	5.56
18	56.25	43.75	18.75	37.50	37.50	6.25
19	58.82	41.18	47.06	23.53	11.76	17.65
20	33.33	66.67	33.33	16.67	0.00	50.00
21	41.67	58.33	30.56	44.44	11.11	13.89
22	60.53	39.47	23.68	23.68	36.84	15.79
Mean	**59.01**	**40.99**	**33.11**	**23.04**	**25.89**	**17.96**
SD	**19.20**	**19.20**	**20.28**	**13.03**	**21.37**	**13.04**

dant recurrences as percentages of total recurrences for each participant. All statistical analyses were performed on these percentages. On average, there were 66.68 (SD = 31.60; Range: 20 - 141) instances per participant where a stutter on a word in a reading was not followed by a stutter on the same word in the next reading (i.e., absence of recurrence). On average, there were 37.36 instances of absence of recurrence for repetitive stutters (SD = 24.58; Range: 9 - 115) and 29.32 for non-repetitive stutters (SD = 17.12; Range: 5 - 68) per participant.

Concordant and Discordant Recurrences

A t-test for related samples between total concordant (a repetitive or non-repetitive stutter on a word in a reading was followed by the same type of stutter in the adjacent reading) and total discordant recurrences showed that there were significantly more concordant than discordant stutters ($t21 = -2.20$, $p = .03$, Cohen's $d = 0.469$). A t-test for related samples between concordant repetitive recurrences (a repetitive stutter on a word in a reading was followed by a repetitive stutter on the same word in the next reading) and discordant repetitive recurrences (a repetitive stutter on a word in a reading was followed by a non-repetitive stutter on the same word in the next reading) was not significant ($t21 = -1.79$, $p = .08$, Cohen's $d = 0.389$) indicating that a repetitive stutter was equally likely to be followed by a repetitive or a non-repetitive stutter. Similarly, a paired-sample t-test between concordant non-repetitive recurrences (a non-repetitive stutter on a word in a reading was followed by a non-repetitive stutter on the same word in the next reading) and discordant non-repetitive recurrences (a non-repetitive stutter on a word in a reading was followed by a repetitive stutter on the same word in the next reading) was not significant ($t21 = -1.33$, $p = 0.19$, Cohen's $d = 0.289$) mirroring the finding for repetitive stutters.

Discussion

With regard to the first research question, whether stutters in general are the direct result of an impairment or adaptive responses to an underlying impairment, the present study appears to suggest that a significant number but, perhaps, not all stutters are the direct result of an impairment. If stutters

were adaptive responses, there would have been significantly more discordant than concordant stutters. Typically, unsuccessful adaptive responses are abandoned in favor of something else. However, the effect size of 0.469 is moderate (Cohen, 1988) and does not account for all of the variability. This suggests that some of the stutters may be compensatory.

The second research question we posed anticipated the possibility that some stutters are the direct result of an impairment and others are compensatory. We hypothesized that repetitive stutters (sound and syllable repetitions), which are characteristic of incipient stuttering, are perhaps the direct result of an impairment and non-repetitive stutters (sound prolongations, broken words, and tense pauses), which are typically the features of chronic stuttering, may be adaptive responses. Unfortunately, the present study did not resolve this question satisfactorily. At p=0.08, concordant repetitive stutters fell just short of being significantly more numerous than discordant repetitive stutters. The effect size of 0.389 is regarded as moderate (Cohen, 1988).

The concordant non-repetitive stutters did not differ from discordant non-repetitive stutters. Non-repetitive stutters are more variable in recurrence and greater variability is a feature of adaptive responses. People who stutter show a variety of "secondary" behaviors such as closing or blinking eyes, wrinkling forehead, stomping foot, etc. Their use of these superfluous behaviors is typically highly variable from one stutter event to the next and they are considered adaptive or maladaptive. Perhaps non-repetitive stutters fall into the same category. Over the years, the core features of stuttering (i.e., stutters that serve as diagnostic markers) have been steadily narrowed now to include what Wingate (1964) referred to as audible or silent repetition of sounds, syllables, and one-syllable words and audible and silent prolongation of sounds. Perhaps a case can be made to further narrow the core to include just the audible sound and syllable repetition of words, which appears to be the universal feature at the onset of stuttering. Everything else we observe in a person who stutters may be an attempt, some habitual and others on-the-fly, to minimize or circumvent this impediment to the forward flow of speech.

Interestingly, one of the most successfully used treatment techniques for stuttering, rate control (Bothe, et al., 2006), involves training clients to use controlled prolongation of syllables as a way of facilitating fluency. It would be difficult to argue that sound prolongation is a stutter and it can also be used as a treatment technique for stuttering. It is more rational to hold that uncontrolled prolongation is a maladaptive response to stuttering while controlled prolongation is a beneficial, adaptive response. A similar explanation may be offered for tense pauses and broken words, which are presumably due to the vocal folds locked in a tense adductive or abductive position at the onset of a word or in the middle of a word respectively. To counteract these maladaptive responses, clients are typically trained to use a breathy onset of voice. It appears that what have been referred to as dysrhythmic phonations (Yairi & Ambrose, 1999) or as audible or silent prolongation of sounds and silent repetition of sounds (Wingate, 1964) are maladaptive responses, which are replaced with controlled and modified versions of the same in certain widely used stuttering treatment programs.

Limitations of the study

Stuttering is highly variable in both time (lifespan and moment-to-moment) and place (communication situations and partners). This presents a serious challenge for its systematic study. Recurrent stutters in repeated readings offer a unique and otherwise unobtainable perspective into the mechanism of stuttering because these stutters occur arguably in the most controlled condition possible. However, recurrent stutters are relatively infrequent and are a function of the frequency of stutters. People with mild to moderate stuttering tend to have very few recurrent stutters. As discussed earlier, the small sample size of the present study may have lacked adequate statistical power to answer adequately all of the research questions posed due to the high degree of variability of stuttering.

In this preliminary study, we analyzed recurrent stutters in the context of two broad types of stutters - repetitive and non-repetitive. Addressing the same research questions with a finer classification of stutters - sound repetitions, syllable repetitions, sound prolongations, tense pauses, and broken words, for instance - using a much larger sample may be able to better discriminate those stutters that are the direct result of an impairment from those that are adaptive responses to an underlying impairment. Although there are numerous neuroimaging studies of PWS and stuttered speech, they have generally resulted in incongruent findings implicating a large number of diverse cortical and subcortical structures and pathways (Ingham, Grafton, Bothe & Ingham, 2012). Properly designed neuroimaging studies of recurrent stutters may provide the most definitive description of the neural mechanism of stuttering. Such studies may elucidate the brain regions and pathways activated by concordant and discordant stutters and may overcome the present quandary in identifying ".... stable neural regions that are consistently associated with stuttering..." (Ingham et al., 2012, p. 12). Occasionally, people with severe stuttering have stutters on the same word in four, five, or even six repeated readings. Some of these recurrent stutters are likely to be concordant and others discordant. Mapping brain regions and pathways involved in these massively recurrent stutters on a single word in nearly identical speaking conditions will enhance our understanding of the brain mechanisms under-

lying stutter events. A comparison of brain regions and pathways of recurrent stutters in a pair of readings with instances of non-recurrence on the same word in another pair of readings will also provide information on brain regions and pathways involved in stutters and fluent speech in PWS.

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