Differential diagnosis in developmental and acquired neurogenic stuttering: Do fluency-enhancing conditions dissociate the two?

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A B S T R A C T

In the past, several authors have proposed comparable sets of clinical features to differentiate acquired neurogenic stuttering (ANS) from developmental stuttering (DS). Further, people with ANS have been reported to show no changes in their dysfluencies during various fluency-enhancing conditions. Although these features have been criticized on their aptness and reliability in differentiating the two disorders (e.g., Lebrun, Bijleveld, & Rousseau, 1990), clinicians and researchers around the world continue to use them even today. In this context, we compile evidence from investigations employing fluency-enhancing conditions in people with ANS to highlight that this group shows extreme variability (including beneficial effects) under such conditions. Further, by combining the evidence from this review as well as that of Lebrun and colleagues', we propose that the clinical features that are used to differentiate ANS from DS are often unreliable. Additionally, we highlight on: (a) the heterogeneity in the manifestation of ANS, (b) recent attempts to draw similarity between ANS and DS, as well as (c) the surprising dearth of functional neuroimaging investigations in ANS that could pave potential ways to future investigations in this fluency disorder.

1. Introduction

Acquired neurogenic stuttering (ANS), as the name signifies, makes a sharp distinction from developmental stuttering (DS) in terms of its onset. The former typically develops with an apparent neurological event (i.e., brain lesion) and is more commonly seen in adults as opposed to DS which gradually develops and progresses from childhood. In addition to this cardinal feature, several other salient features of ANS have been proposed by various authors. For instance, Manning (2001) listed out six clinical features that are often used to differentiate ANS from DS. These include: (i) roughly equal occurrence of dysfluencies on both content and function words; (ii) lack of anxiety about dysfluencies; (iii) stuttering instances not restricted to initial syllables; (iv) rare occurrence of secondary behaviors; (v) absence of adaptation effect; and (vi) occurrence of dysfluencies on all speech tasks. Several authors have reported of comparable sets of features to differentiate ANS from DS (e.gs., Helm-Estabrooks, 1999; Lundgren, Helm-Estabrooks, & Klein, 2010; Ringo & Dietrich, 1995; Rosenbek, Messert, Collins, & Wertz, 1978). It may be noted that the last two features reported by Manning (2001) (and third & sixth features in Lundgren et al.'s (2010) review) are related to the fluency-enhancing conditions. However, in 1990, Lebrun, Bijleveld, and Rousseau published a critical review of clinical features of ANS (see below) and argued that the commonly used differential diagnostic features of ANS are often fallible and unreliable. Yet, Lebrun and colleagues' proposal has largely been ignored as subsequent reports have continued to adhere to the differential features of ANS mentioned above. In this context, we confirm the arguments of Lebrun et al. by reviewing the disparate evidence, primarily from the fluency-enhancing conditions in an attempt to draw the attention of speech language pathologists and neuroscientists to the fallible and unreliable nature of differential diagnostic features of ANS. Before elucidating such evidence, in the following section, we briefly present the excerpt of Lebrun and colleagues' review.

2. Lebrun and colleagues' critical review

In their review, Lebrun et al. (1990) presented several studies that showed features discordant to the proposed differential diagnostic features of ANS. For instance, from the observations of Lebrun and Van Borsel (1990), they challenged the belief that the word-final dysfluencies are uncommon in DS. Further, the authors argued that people with ANS could show only word-initial dysfluencies (e.gs., Rosenbek, McNeil, Lemme, Prescott, & Alfrey, 1978; Rosenbek, Messert, et al., 1978). In terms of the speech units that are stuttered, Lebrun and Leleux (1985) opposed the belief that people with ANS repeat relatively larger segments such as phrases or sentences. Similarly, they observed that the parts of speech (e.g., content and function words) do not reliably differentiate ANS from DS. With respect to the speech tasks (e.g., spontaneous speech, reading, & singing), they garnered evidence that refutes the assumption that only people with DS show dysfluencies in tasks that require more spontaneity (e.g., spontaneous speech as opposed to choral speech or oral reading). To support their argument Lebrun et al. cited the case of a person with ANS (Baratz & Mesulam, 1981) who exhibited stuttering only during spontaneous speech, with apparently fluent oral reading. Similarly, evidence for task-based variations in dysfluencies was secured from the investigations of Mazzuchi and colleagues (Mazzuchi, Moretti, Carpeggiani, Parma, and Paini, 1981), Lebrun and Leleux (1985), and Lebrun, Devreux, and Rousseau (1986) by these authors. Thus, with these observations, Lebrun et al. (1990) refuted the clinical features that are often used to differentiate ANS from DS. In the following section, we compile further evidence on the fallibility of fluency-enhancing conditions in the differential diagnosis of ANS from DS.

3. ANS and fluency-enhancing conditions

Several conditions have been proposed to eliminate dysfluencies in the speech of people with DS. These include: altered (i.e., masked [MAF], delayed [DAF], or frequency altered [FAF]) auditory feedback, singing, choral (or unison) speech, and repeated reading (i.e., adaptation effect). Quite often, lack of reduction in dysfluencies under these conditions has been reported as salient to ANS (e.gs., Lundgren et al., 2010; Manning, 2001). In the following section, we present a brief review of some studies demonstrating an ameliorative effect of fluency-enhancing conditions for ANS.

In a recent study, Krishnan and Tiwari (2011) reported the case of a 56-year-old lady with ANS who exhibited marked reduction in dysfluencies under MAF. Their observation was in sharp contradistinction to the salient clinical feature of ANS that the dysfluencies occur in all speech tasks (e.g., Manning, 2001). Similarly, Van Borsel, Drummond, and Pereira (2010) provided evidence for fluency enhancement under DAF from a 49-year-old patient with ANS. They observed a general reduction of dysfluencies with 70 ms DAF compared to 110 ms delay in reading aloud and in spontaneous speech tasks. In addition to the evidence from recent investigations, researchers in the past have also reported of beneficial effect of DAF in ANS (e.gs., Marshall & Neuburger, 1987; Marshall & Starch, 1984). Together from their studies, Marshall and colleagues reported of marked reduction in dysfluencies with a delay of 250 ms in four people with ANS. Similarly, Downie, Low, and Lindsay (1981) reported of a 60-year-old man with Parkinson's disease who showed remarkable improvement in speech fluency with 50 ms delay in auditory feedback. These authors reported on yet another subject with ANS who showed positive changes in fluency only for about a year. Though these studies have demonstrated beneficial effects of DAF and MAF in ANS, several studies failed to show such effects. For instance, Balasubramanian, Max, Van Borsel, Rayca, and Richardson (2003) reported of a subject (CP) who failed to show reduction of dysfluency under FAF and DAF. Further, Jokel, De Nil, and Sharpe (2007) did not find any improvement in fluency under masking noise in a group of 12 persons with ANS following either stroke or trauma to the brain. Similarly, in yet another study, Balasubramanian, Cronin, and Max (2010) reported of two subjects who failed to show improvement in fluency under DAF. Rather, in their subjects, the authors reported of an increment in dysfluencies under AAF. For instance, in their first subject, no apparent variation in stuttering under DAF compared to normal auditory feedback was observed. However, in the same patient, shifting the auditory feedback either by half octave up or down resulted in an increment in dysfluencies. The second subject, on the other hand, failed to show dysfluency reduction under both DAF and FAF conditions. Balasubramanian et al. (2010) speculated that these differential performances were possibly due to individual differences in the underlying pathology, which could affect the availability of intact neural resources that in turn could be recruited in order to benefit the fluency-enhancing conditions.

Singing is a universal task that facilitates speech fluency in people with DS (Wan, Rüber, Hohmann, & Schlaug, 2010). According to the last criterion of Manning (2001) that differentiates ANS from DS (and third of Lundgren et al., 2010), signing is unlikely to facilitate fluency in people with ANS. However, in their review, Lebrun et al. opposed this argument with evidence from several investigations (e.gs., Lebrun et al., 1986; Lebrun & Leleux, 1985). Similarly, in a recent investigation, Krishnan and Tiwari (2011) reported of reduction of dysfluencies while singing in a person with ANS. In Theys and colleagues' (Theys, van Wieringen, & De Nil, 2008) survey, majority of the respondents (speech-language therapists) reported to have observed beneficial effect of singing in people with ANS. Thus, it is apparent from this mixture of recent and past investigations that people with ANS may show marked reduction of dysfluencies while singing.

Yet another fluency-enhancing condition that is acclaimed to bring in quick and radical reduction in dysfluencies of people with DS is choral (or unison) speech (Freeman & Armson, 1998; Saltuklaroglu, Kalinowski, Robbins, Crawcour, & Bowers, 2009). In this task, persons with stuttering simultaneously produce the utterances with clinician. According to the sixth differential diagnostic feature compiled by Manning (see Para. 1), this task is believed to have trifling effect on fluency enhancement in people with ANS. Yet, the available studies on choral reading in ANS show mixed results. For instance, while Jokel et al. (2007) reported of improved speech fluency with choral speech, Balasubramanian et al. (2010) did not observe any such effect in their subjects with ANS.

The adaptation effect – reduction of dysfluencies while repeatedly reading the same text (Johnson & Knott, 1937) – is often noticed in people with DS. The absence of this effect has been a salient feature of ANS (Lundgren et al., 2010; Manning, 2001). Interestingly, recent investigations have shown that the adaptation effect is highly variable in people with ANS. For instance, Krishnan and colleagues (Krishnan, Nair, & Tiwari, 2010) reported of adaptation effect in their 51-year-old subject with ANS. Similarly, Tani and Sakai (2011) observed this effect in a series of five subjects with ANS. Supportive observations were also reported by Balasubramanian et al. (2003) and Theys et al. (2008). Further, in Theys and colleagues' survey, 19% of 52 clinicians claimed to have observed adaptation effect in people with ANS. Yet, some studies have not observed this effect (e.gs., Balasubramanian et al., 2010; Krishnan

& Tiwari, 2011) in people with ANS. Thus, the recent investigations show mixed findings on adaptation effect in ANS, making it an unreliable differential diagnostic feature.

In the context of growing evidence on the frailty of various fluency-enhancing conditions, a novel measure to differentiate ANS from DS is noteworthy. For instance, using a combinatorial (i.e., perceptual & acoustic-spectrographic) analysis, Viswanath (2009–2010) claimed that people with ANS produce significantly more inter- than intra-syllabic repetitions, whereas those with DS showed the opposite pattern. Further, the duration of repeated vowels during intra-syllabic repetitions is significantly longer in ANS compared to DS. Finally, based on the observations, Viswanath proposed that people with ANS produce fully elaborated target vowels during part-word repetitions, rather than schwa vowels, unlike those with DS who show the opposite pattern. Yet, it may be noted that more investigations are necessary to elucidate the aptness of part-word repetitions in the differential diagnosis of ANS from DS.

From the brief review above, it is apparent that people with ANS showed marked heterogeneity in their performance under fluency-enhancing conditions. Thus, the reliance on these features to differentiate ANS from DS may lead to misleading clinical decisions. Further, our observations support the views of Lebrun et al. (1990) that the existing clinical behavioral dissociations are frailer and unreliable. In this context, the analysis of part-word repetitions seems to be a promising approach to differentiate ANS and DS, though it requires further validation. We now briefly discuss the heterogeneous neural substrates of ANS in the following section.

4. Neural substrates of ANS

The investigators of ANS are often bewildered by a myriad of lesion sites in the brain that leads to this speech disorder. Virtually, every lobe of the brain, including occipital (Case 4: Grant, Biousse, Cook, & Newman, 1999), has been implicated with ANS. Further, ANS has been reported in people with lesions to corpus callosum (Hamano et al., 2005), brainstem (Balasubramanian et al., 2003), and cerebellum (Tani & Sakai, 2010). Furthermore, distinct etiologies such as stroke (Grant et al., 1999; Jokel et al., 2007), traumatic brain injury (Helm-Estabrooks & Hotz, 1998; Lebrun et al., 1990), seizure disorder (Sechi, Cocco, D'Onofrio, Deriu, & Rosati, 2006), encephalitis (Chen & Peng, 1993), Parkinson's disease (Goberman, Blomgren, & Metzger, 2010), and dementias (Quinn & Andrews, 1977; Rosenbek, McNeil, Lemme, Prescott, & Alfrey, 1978) have been associated with ANS. The diversity in etiology, and more importantly, the lesion sites adumbrates the complexity in the neural substrates of fluent speech production. Accordingly, some people with ANS show similarity in their clinical profile to that of people with DS. In this context, we next review a recent investigation that attempted to draw parallels between ANS and DS.

5. Novel directions in the investigations on ANS

In 2011, Krishnan and Tiwari argued that it may be productive and promising to find similarities between ANS and DS, rather than differentiating the two disorders. They based this argument on their observations of a person with ANS under various fluency-enhancing (and also under a treatment technique – pacing) conditions. These authors could explain the performance of their subject under the fluency-enhancing conditions in the light of the recent explanatory hypotheses of DS (for details, see Krishnan & Tiwari, 2011). In the context of rising evidence on the clinical features that fail to distinguish ANS from DS, such attempts to find similarity between these two fluency disorders may be constructive, as they may provide novel insights into the mechanism of dysfluent speech in the these disorders, as well as to extend the management strategies (e.g., Krishnan & Tiwari, 2011) from DS to ANS.

Finally, based on Krishnan and Tiwari's (2011) observation on the lack of functional neuroimaging investigations in ANS, we reiterate the need to extend the use such imaging techniques from DS to ANS on two accounts. First, application of functional neuroimaging techniques may fill the gap between these disorders, especially at the neural level. Second, these techniques may be used to track the functional changes in the brain associated with (treatment-related) recovery, which, in turn, help us to refine the neural underpinnings of fluent speech production.

6. Summary

Here, we have shown that even after two decades of an earlier critique, ANS prevails with a set of differential diagnostic features that are often fallible and unreliable. Hence, the heterogeneity in lesion sites and types as well as in the clinical profile including those under various fluency-enhancing conditions are actually, more the norm in ANS. The recent attempts to emphasize similarities between ANS and DS, rather than differences, may be more promising in the future.

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References

Balasubramanian, V., Cronin, K. L., & Max, L. (2010). Dysfluency levels during repeated readings, choral readings, and readings with altered auditory feedback in two cases of acquired neurogenic stuttering. *Journal of Neurolinguistics, 23*(5), 488–500.
Balasubramanian, V., Max, L., Van Borsel, J., Rayca, K. O., & Richardson, D. (2003). Acquired stuttering following frontal and bilateral pontine lesion: a case study. *Brain and Cognition, 53*, 185–189.
Baratz, R., & Mesulam, M. (1981). Adult-onset stuttering treated with anticonvulsants. *Archives of Neurology, 38*, 132.
Chen, W.-H., & Peng, M.-C. (1993). Acquired stuttering in a patient with encephalitis. *Kaohsiung Journal of Medical Sciences, 9*, 183–185.
Downie, A. W., Low, J. M., & Lindsay, D. D. (1981). Speech disorder in Parkinsonism: use of delayed auditory feedback in selected cases. *Journal of Neurology, Neurosurgery, & Psychiatry, 44*, 852–853.
Freeman, K., & Armson, J. (1998). Extent and stability of stuttering reduction during choral reading. *Journal of Speech-Language Pathology & Audiology, 22*, 188–202.
Goberman, A. M., Blomgren, M., & Metzger, E. (2010). Characteristics of speech fluency in Parkinson disease. *Journal of Neurolinguistics, 23*(5), 470–478.
Grant, A. C., Biousse, V., Cook, A. A., & Newman, N. (1999). Stroke-associated stuttering. *Archives of Neurology, 56*, 624–627.
Hamano, T., Hiraki, S., Kawamura, Y., Hirayama, M., Mutoh, T., & Kuriyama, M. (2005). Acquired stuttering secondary to callosal infarction. *Neurology, 64*, 64–65.
Helm-Estabrooks, N. (1999). Stuttering associated with acquired neurological disorders. In R. F. Curlee (Ed.), *Stuttering and related disorders of fluency* (2nd ed.). (pp. 255–268) New York: Thieme.
Helm-Estabrooks, N. A., & Hotz, G. (1998). Sudden onset of stuttering in an adult: neurogenic or psychogenic? *Seminars in Speech and Language, 19*, 23–29.
Johnson, W., & Knott, J. R. (1937). Studies in the psychology of stuttering: I. The distribution of moments of stuttering in successive readings of the same passage. *Journal of Speech and Hearing Disorders, 2*, 17–19.
Jokel, R., De Nil, L. F., & Sharpe, A. K. (2007). Speech disfluencies in adults with neurogenic stuttering associated with stroke and traumatic brain injury. *Journal of Medical Speech-Language Pathology, 15*(3), 249–257.
Krishnan, G., Nair, R. P., & Tiwari, S. (2010). Clinical evidence for the compensatory role of right frontal lobe and a novel neural substrate in developmental stuttering: a single case study. *Journal of Neurolinguistics, 23*, 501–510.
Krishnan, G., & Tiwari, S. (2011). Revisiting the acquired neurogenic stuttering in the light of developmental stuttering. *Journal of Neurolinguistics, 24*, 383–396.
Lebrun, Y., Bijleveld, H., & Rousseau, J.-J. (1990). A case of persistent neurogenic stuttering following a missile wound. *Journal of Fluency Disorders, 15*, 251–258.
Lebrun, Y., Devreux, F., & Rousseau, J.-J. (1986). Language and speech in a patient with a clinical diagnosis of progressive supranuclear palsy. *Brain and Language, 27*, 247–256.
Lebrun, Y., & Leleux, C. (1985). Acquired stuttering following right brain damage in dextrals. *Journal of Fluency Disorders, 10*, 137–141.
Lebrun, Y., & Van Borsel, J. (1990). Final sound repetitions. *Journal of Fluency Disorders, 15*, 107–113.
Lundgren, K., Helm-Estabrooks, N., & Klein, R. (2010). Stuttering following acquired brain damage: a review of the literature. *Journal of Neurolinguistics, 23*(5), 447–454.
Manning, W. (2001). *Clinical decision-making in fluency disorders*. Vancouver: Singular/Thompson Learning.
Marshall, R. C., & Neuburger, S. I. (1987). Effects of delayed auditory feedback on acquired stuttering following head injury. *Journal of Fluency Disorders, 10*, 241–255.
Marshall, R. C., & Starch, S. A. (1984). Behavioral treatment of acquired stuttering. *Australian Journal of Human Communication Disorders, 12*, 87–92.
Mazzuchi, A., Moretti, G., Carpeggiani, P., Parma, M., & Paini, P. (1981). Clinical observations on acquired stuttering. *British Journal of Disorders of Communication, 16*, 19–30.
Quinn, P. T., & Andrews, G. (1977). Neurological stuttering: a clinical entity. *Journal of Neurology, Neurosurgery, & Psychiatry, 40*, 699–701.
Ringo, C. C., & Dietrich, S. (1995). Neurogenic stuttering: an analysis and critique. *Journal of Medical Speech-Language Pathology, 3*, 111–122.

Rosenbek, J., McNeil, M. R., Lemme, M. L., Prescott, T. E., & Alfrey, A. C. (1978). Speech and language findings in a chronic hemodialysis patient: a case report. *Journal of Speech and Hearing Disorders, 40*, 245–252.
Rosenbek, J., Messert, B., Collins, M., & Wertz, R. T. (1978). Stuttering following brain damage. *Brain and Language, 6*, 82–96.
Saltuklaroglu, T., Kalinowski, J., Robbins, M., Crawcour, S., & Bowers, A. (2009). Comparisons of stuttering frequency during and after speech initiation in unaltered feedback, altered auditory feedback and choral speech conditions. *International Journal of Language & Communication Disorders, 44*(6), 1000–1017.
Sechi, G. P., Cocco, G. A., D'Onofrio, M., Deriu, M. G., & Rosati, G. (2006). Disfluent speech with partial epilepsy: beneficial effects of levetiracetam. *Epilepsy & Behavior, 9*, 521–523.
Tani, T., & Sakai, Y. (2010). Stuttering after right cerebellar infarction: a case study. *Journal of Fluency Disorders, 35*, 141–145.
Tani, T., & Sakai, Y. (2011). Analysis of five cases with neurogenic stuttering following brain injury in the basal ganglia. *Journal of Fluency Disorders, 36*(1), 1–16.
Theys, C., van Wieringen, A., & De Nil, L. F. (2008). A clinician survey of speech and non-speech characteristics of neurogenic stuttering. *Journal of Fluency Disorders, 33*, 1–23.
Van Borsel, J., Drummond, D., & Pereira, M. M. B. (2010). Delayed auditory feedback and acquired neurogenic stuttering. *Journal of Neurolinguistics, 23*(5), 479–487.
Viswanath, N. S. (2009–2010). Part-word repetitions by persons with acquired and developmental stuttering: a pilot spectrographic investigation. *Texas Journal of Audiology and Speech-Language Pathology, XXXII*, 12–16.
Wan, C. Y., Rüber, T., Hohmann, A., & Schlaug, G. (2010). The therapeutic effects of singing in neurological disorders. *Music Perception, 27*(4), 287–295.