Stuttering: Imagining a Solution to the Riddle

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Abstract

The old saying by Van Riper (with a nod to Winston Churchill), that stuttering is a riddle wrapped in a mystery inside an enigma, is well known. While it would be preposterous for any of us to state that, 40 years later, we now have solved the riddle, we can say with a great degree of confidence that advances in scientific inquiry have brought us closer to understanding the factors that may trigger the onset, development, and/or maintenance of stuttering. Nevertheless, much still needs to be learned, because the riddle still poses many challenges. For instance, we do not fully understand why developmental stuttering starts somewhere between 2 and 9 years of age, but onset after puberty is rarely if ever seen (other than neurogenic stuttering, but that is a different story), or why boys are more likely to develop chronic stuttering than girls are.

Another unsolved question is the variability seen in stuttering severity. Individuals who stutter often will report having more stuttering in some situations than others, or when talking to some listeners, or even simply from day to day. Similarly, someone with very severe stuttering typically will not stutter on more than approximately 40% of their words. So, what happens with or during the other 60%? And while we have learned a lot about treatment efficacy for children and adults who stutter, we still cannot explain why some clients do well in maintaining their fluency following treatment, while others relapse.

These are only a few of the questions that remain to be answered. Over the past 10 to 15 years, my colleagues and I slowly have chipped away at some of the facets of the riddle, as have many other researchers across the world. In this article, I will share some of our findings and the tentative conclusions we have drawn so far. In the following sections, I will review three main themes in our research: (a) people who stutter have a reduced ability to incorporate proprioceptive feedback in their movement execution, (b) people who stutter demonstrate difficulty acquiring new motor skills, and (c) behavioural differences in sensory and motor skills between stuttering and nonstuttering individuals may be related to distinctions in their functional and structural organization of the cortical and subcortical processes.

The Role of Proprioceptive Feedback in Stuttering

One of the central tenets in the research efforts of my colleagues and I has been that stuttering can be understood best as a speech motor disorder (De Nil, 1999a) while still acknowledging the potential influence of language (Sasisekaran & De Nil, 2006), psychological
factors such as temperament (Eggers, De Nil, & van Den Bergh, in press), or other influences in triggering and/or affecting the further development of speech fluency disruptions in both adults and children. In our earlier work, we focused primarily on the role of proprioceptive feedback (the feedback arising from muscles, tendons, joints, and skin sensory organs that allows us to know the location and movement of our body in space) in the control and coordination of speech movements. As a result, we were able to demonstrate that adults who stutter are less proficient than nonstuttering speakers in using proprioceptive feedback for movement control regardless of whether these movements speech-related or nonspeech-related. For instance, De Nil and Abbs (1991) showed that adults who stutter were less able to use proprioceptive feedback for controlling the amplitude of very small jaw, lip, and tongue movements. Compared to nonstuttering controls, their movements, on average, were twice as imprecise. Interestingly, the same was true for finger movements, suggesting that this sensory deficit may not be limited to oral or speech-related movements. We also found that the ability to make small movements significantly was correlated with stuttering severity, but with a surprising twist (Archibald & De Nil, 1999): Only participants with mild stuttering, and not those with moderate or severe stuttering, performed poorer on a proprioceptive minimal movement task (making ‘smallest possible’ jaw movements in the absence of visual feedback) compared to typically speaking controls. The more severe stuttering subjects, however, had significantly slower movements than any of the other two groups (i.e., mild stuttering, moderate stuttering). This suggested that when relying on proprioceptive feedback, those with more severe stuttering tended to slow down their movements in order to improve accuracy, and as a result did indeed perform better.

As part of his doctoral dissertation, Torrey Loucks extended this earlier work by showing that the relatively reduced proprioceptive sensitivity in adults who stutter resulted in poorer performance on oral motor tasks that involved precise target oriented movements of the jaw (Loucks, De Nil, & Sasisekaran, 2007), which became even less precise under time pressure (Loucks & De Nil, 2006b). Reduced proprioceptive resolution also has been found to reduce stuttering individuals’ ability to coordinate jaw movements with phonation, suggesting that it may be a key factor in the observed articulatory discoordinations often observed in people who stutter (Loucks et al., 2007). The observations from these and other studies have raised a number of interesting questions. First of all, the finding that severe stuttering speakers adjusted their movement strategy to increase accuracy of movement may suggest that at least some of the observed differences in the use of proprioceptive feedback could have been driven by movement strategy rather than sensory sensitivity. To test this hypothesis, we conducted a masseter muscle vibration experiment (Loucks & De Nil, 2006a). Vibration excites the proprioceptive sensors, primarily those located in muscle tendons, and results in an involuntary illusion that muscles are shortening, and thus that the articulators are moving, while in fact they are not or at least not as much as perceived. As expected, our results showed that both stuttering and nonstuttering individuals were affected by this illusion, but the illusion was stronger for the nonstuttering adults, suggesting that the stuttering speakers indeed had a decreased proprioceptive sensitivity. Therefore, our movement data most likely reflected underlying physiology rather than movement strategy. A second related question is whether the increase in movement accuracy associated with a slowing of movement speed, as reported by Archibald and De Nil (1999), can be linked to the observation that stuttering speakers become more fluent when slowing their rate of speech. It is possible that this fluency enhancing effect of slowed speech is at least partly the result of improved proprioceptive feedback. We do not yet have data to answer this question which requires further investigation.

**Motor Skill Acquisition**

A second line of research in our lab has been to investigate whether stuttering speakers find it harder to acquire new motor skills compared to nonstuttering speakers. Accurate sensory input is a crucial component of motor learning, allowing us to plan and guide our
movements within the context of the desired target and environmental conditions. It also allows
for appropriate error correction if the actual versus intended outcomes are (too) far apart. As
we have discussed in the previous paragraph, our research has demonstrated that people who
stutter have difficulty with tasks that require fine proprioceptive feedback.

Proprioceptive feedback plays an important role in speech production. Research has
shown that such feedback allows us to quickly and often unconsciously compensate for
perturbations of our articulatory movements (Namasivayam, van Lieshout, & De Nil, 2008).
Proprioception is also important for the spatial and temporal coordination of fluent articulatory
movements, a fact that did not go unnoticed for Van Riper, who included proprioceptive
training as an important component of fluency treatment (Van Riper, 1982). Learning a new
motor skill relies heavily on the availability of accurate and fast proprioceptive feedback
(Schmidt & Lee, 1998). Such afferent information is integrated quickly and smoothly with
efferent motor signals to guide movements toward the desired target outcome. Neurological
disorders which negatively affect afferent proprioceptive information typically result in motor
learning deficits (Bard, Fleury, Teasdale, Paillard, & Nougier, 1995; Messier, Adamovich,
Berkinblit, Tunik, & Poizner, 2003). While stuttered speech is characterized by a breakdown in
articulatory coordination, research has shown that even fluent speech in persons who stutter
is not always well coordinated (Max & Gracco, 2005; McClean & Tasko, 2004), suggesting the
presence of a more generalized motor deficit.

In one of the studies previously discussed (Loucks et al., 2007) in which adults who
stutter were provided practice in reaching a specified spatial target with their jaw, we
unintentionally observed that many of them had difficulty, in comparison to nonstuttering
controls, in reaching the a priori specified criterion of 3 consecutive movements within a pre-
specified error range. This observation was one of the reasons that led us to examine speech
and nonspeech motor learning in adults who stutter. Studies completed in our lab have
demonstrated consistently that adults who stutter have difficulty with the acquisition of novel
speech and nonspeech movement skills (Smits-Bandstra, De Nil & Rochon, 2006; Smits-
Bandstra, De Nil & Saint-Cyr, 2006). Sequence skill learning can be defined operationally as
increased efficiency (speed and accuracy) in the initiation and completion of individually known
movements combined to form a novel movement sequence over practice. Humans effortlessly
perform complex, coordinative movements almost constantly during everyday life. In order to
do this effectively, people must minimize the need for conscious effort and online control by
making the transition from an attention-focused movement control strategy to a relatively
automatic movement pattern. Brain studies have confirmed the presence of distinct patterns of
brain activity when motor sequence learning is mediated by attention (e.g., in early stages of
learning) compared to when attention to motor performance is minimal (Keele, Mayr, Ivry,
Hazeltine, & Heuer, 2003).

The basic paradigm in our studies consisted of asking participants to complete a
relatively simple but novel movement sequence repeatedly during a single session. This
movement sequence consisted either of speech (syllable string) or of nonspeech (finger tapping)
sequences. In general, adults who stutter were slower in learning these patterns. This group
difference especially was apparent in the initiation of the movements, but also could be seen in
sequence duration, resulting in a less steep learning curve (Smits-Bandstra, De Nil & Rochon,
2006; Smits-Bandstra, De Nil & Saint-Cyr, 2006). In a more recent study, we have shown that
this difference continues following more extensive practice and consolidation over multiple days
(Bauerly & De Nil, 2009).

Differences between stuttering and nonstuttering speakers also have been observed
during a dual task learning paradigm (Smits-Bandstra, De Nil, & Rochon, 2006; Smits-
Bandstra & De Nil, 2009). While a competing task will interfere with motor learning, its
disruptive effects will become less pronounced as the primary task becomes more automated
because it requires less cognitive control. For speech in particular, given the need for rapid and
accurate coordination of multiple articulatory movements, a high degree of automaticity is required for the production of fluent speech. Again, our studies have demonstrated that adults who stutter differ from nonstuttering controls in their ability to transition a novel speech and nonspeech sequence to an automatic state. In particular, participants were asked to practice syllable and finger tapping sequences while alternating between a single and dual task mode. In the dual task mode, the visually presented letters and numbers for the speech and tapping task, respectively, changed color repeatedly and the participants were instructed to monitor whether any of the colors were repeated. As expected, the speed of initiating the movement sequences decreased significantly under the dual task condition for the nonstuttering speakers. To our surprise, no such effect was observed for the stuttering speakers. Instead, their performance remained relatively constant, but at the level of the dual task performance for the control participants. We interpreted these findings in two ways. First, the data suggested that the stuttering participants consistently performed the speech and tapping task at a ‘dual task’ level, indicating the need for continued greater conscious effort and voluntary control during movement execution. Unlike the performance of nonstuttering participants, their performance did not appear to transition to a more automatic, less controlled, movement pattern. Second, group differences were found for performance accuracy in the movement and color memory task. Stuttering subjects made more sequencing errors under the dual vs. the single task condition compared to the nonstuttering control subjects. On the other hand, the accuracy in identifying whether colors were repeated or not was not affected significantly by the single vs. dual task conditions. This difference between accuracy for sequence completion and the competing color task suggests that the stuttering participants tended to focus more on the competing color task and on maintaining movement initiation and completion speed, even if this meant sacrificing movement accuracy. If true, this may account in part for the relative lack of dual task interference in movement speed in the stuttering group. However, this hypothesis definitely deserves further investigation.

**Brain Imaging**

A third area of investigation for us has been the use of functional and structural imaging tools to investigate cortical and subcortical brain differences between stuttering and nonstuttering individuals. In a recent publication, we have argued that the findings of deficient motor learning and automaticity capacity in people who stutter may provide indirect evidence for the presence of a dysfunction in cortico-striato-thalamo-cortical connections, and that this may be one etiological component in the development and maintenance of stuttering (Smits-Bandstra & De Nil, 2007). Of course, behavioral observation and analysis only can provide indirect evidence for involvement of specific neural systems in stuttering. Fortunately, brain imaging tools are now widely available for researchers to investigate neural function more directly.

Since the mid 1990s, a growing number of functional and structural brain imaging studies have been conducted in an attempt to unravel the neurological bases of developmental stuttering. While some of the earlier studies were done using positron emission tomography (PET), most current studies have been based on functional magnetic resonance imaging (fMRI), or magnetoencephalography (MEG). In a review of these studies (De Nil, 2004), we concluded that the findings available at that time pointed to two main conclusions. The first one was the observation of overactivation (compared to nonstuttering speakers) of cortical and subcortical (especially cerebellar) areas which have a role in the sensorimotor planning and execution of speech. Second, consistent significant differences between stuttering and nonstuttering speakers were observed in the auditory cortex, primarily characterized by a decrease in functional activation during natural speech. This latter observation suggested that differences between stuttering and nonstuttering speakers involve not only brain regions responsible for motor planning and execution, but also regions which have a role in the processing of sensory feedback. This finding is consistent with some of our behavioral results discussed earlier. A
number of subsequent studies have largely supported these earlier conclusions. For example, Brown, Ingham, Ingham, Laird, and Fox (2005) analyzed data reported in 8 published functional imaging studies that involved both stuttering and nonstuttering adults. They concluded that stuttering speakers (a) overactivate vocal-motor areas, especially in the right hemisphere, (b) activate additional areas with particular involvement of the frontal operculum (RFO) and the cerebellum (vermis), and (c) show a relative absence of bilateral auditory and basal ganglia activations.

As part of our research, we have investigated brain activation changes associated with behavioural fluency treatment through the use of PET (De Nil, 1999b; De Nil & Kroll, 1995; De Nil & Kroll, 2001; De Nil, Kroll, & Houle, 2001; De Nil, Kroll, Lafaille, & Houle, 2003; De Nil, Kroll, McIntosh, & Houle, 1999; Kroll, De Nil, Kapur, & Houle, 1997). Our findings revealed a significantly higher activation during overt and covert word reading prior to treatment in the frontal and medial cortex, and the cerebellum, which became even more pronounced immediately following treatment. After one year of maintenance, this activation became more normalized, showing a sharp reduction in these areas, as well as a more typical left lateralization of function. We interpreted these follow-up findings as being suggestive of an increased level of automaticity during speech production, reduced anticipatory need to scan words for potential stuttering, increased emphasis of on-line self-monitoring, and optimized sequencing and timing of articulatory, phonatory and respiratory movements gained through fluency shaping therapy. Similar results were reported by Neumann, et al. (2003; 2005) who scanned stuttering adults before and immediately after a three-week intensive fluency shaping therapy course, and again after a 2-year follow-up period (for five of the stuttering participants). In contrast to our findings, however, Neumann and her colleagues observed somewhat greater activation in left precentral, middle frontal, anterior cingulate cortex, insula, and putamen regions, and bilaterally in the temporal cortex at the 2-year follow-up period. This difference between the two studies may point to the effects of neuroplasticity associated with ongoing speech skill practice over a longer period of time post-treatment. Also, it will be interesting to see whether interindividual differences in plasticity can be linked to maintenance and relapse of fluency.

One relatively consistent observation in brain imaging of adults who stutter has been a pronounced deactivation of the auditory cortex during speech (Braun et al., 1997; De Nil, Kroll, Kapur, & Houle, 2000; De Nil et al., 2008; De Nil et al., 2003; Fox et al., 1996; van Borsel, Sierens, & Pereira, 2007). Fluency enhancing conditions, such as choral reading, appear to increase auditory activation (Braun et al., 1997; Fox et al., 1996). This observation of reduced auditory activation in stuttering adults has generated significant attention and has led some to suggest that it may result from increased inhibition of auditory cortex functions during overt speech (Brown et al., 2005). Both animal (Eliades & Wang, 2003) and human (Houde, Nagarajan, Sekihara, & Merzenich, 2002) studies have demonstrated inhibition of the primary auditory cortex by motor cortex activation. In one of our recent magnetoencephalography studies, we found that, while both stuttering and nonstuttering adults did show the expected auditory inhibition during overt speech production, the two groups did not differ with respect to the strength of this inhibition effect (Beal, Cheyne, & De Nil, 2007). An alternative explanation may be that redirected attention resources in adults who stutter, who are known to scan for potential stuttering in upcoming speech based on articulatory effort and difficulty, may focus their afferent information processing on the proprioceptive system relative to the auditory system. This may account partially for the consistently observed overactivation of the sensorimotor cortex and deactivation of the auditory cortex during habitual speech. As such, the lower auditory activation in stuttering individuals may not be so much the result of an active inhibition, but rather of a relative lack of sufficient activation during speech. We will test this hypothesis in the near future.
The observation that adults with developmental stuttering show differences in the functional activation of cortical and subcortical regions of the brain leads one to wonder whether these differences may be caused not only by differences in the underlying physiology but possibly also by structural brain differences. The answer to this question now appears to be affirmative as differences have been observed in the volume, density, and gyral patterns of the cortical layers of the lateral motor and auditory cortex, as well as the underlying white matter (Cykowski et al., 2008; Foundas, Bollich, Corey, Hurley, & Heilman, 2001; Foundas et al., 2003; Sommer, Koch, Paulus, Weiller, & Buchel, 2002). We have used voxel based morphometry to study a large group of stuttering and nonstuttering subjects (Beal, Gracco, Lafaille, & De Nil, 2007). Our findings revealed significant gray as well as white matter between-group differences. The most prominent differences were located in the primary auditory cortical region (middle as well as superior temporal gyrus), and more so in the right hemisphere. In addition, increased gray matter volume was found in the left inferior frontal gyrus, including the precentral gyrus and insula, and the right cerebellum. In addition to gray matter differences, the stuttering subjects also showed increased volume in the white matter underlying the right frontal cortex (inferior frontal gyrus, insula and precentral gyrus), as well as the left middle temporal gyrus. Interestingly, these white matter differences were observed in contralateral homologue areas relative to the differences in gray matter. While the significance of this finding is not yet clear, it is possible that it reflects a process by which the contralateral region compensates for a cortical or subcortical structural deficit.

The possible role of specific brain regions in the onset and maintenance of developmental stuttering is supported further by the observation that adults may develop stuttering-like behavior (often referred to as neurogenic or acquired stuttering) following brain lesions or the onset of neurodegenerative disease (De Nil, Jokel, & Rochon, 2007). Neurogenic stuttering is most commonly observed following a stroke (Theys, Vanwieringen, & De Nil, 2008). We have collected extensive behavioral data on a large group of adult stroke patients and are currently in the process of collecting detailed structural and functional brain imaging data as well. Our preliminary data, correlating stuttering severity with the locations of brain lesions, suggests the primary involvement of basal ganglia and premotor cortex, which supports the hypothesis that a dysfunction in this cortical-subcortical system is involved in the control of speech fluency (Alm, 2004; Smits-Bandstra & De Nil, 2007).

Clearly, the data available to date suggests the presence of anatomical and morphological differences between stuttering and nonstuttering adults. To what extent these differences point to etiological versus acquired characteristics remains an open question. First of all, all of the brain imaging studies to date have involved older children and adults, leaving open the possibility that the observed changes result from, rather than trigger, the presence of stuttering. Secondly, and related to the first point, at least some observed differences may reflect self-initiated coping behaviors in adults who stutter. Data from a recent study illustrates how voluntary modifications to speech behavior may differentiate between stuttering and nonstuttering adults (De Nil et al., 2008). We asked a group of stuttering and nonstuttering speakers to speak in three different manners: (1) using their habitual manner; (2) while simulating stuttering (e.g. f..f..f..f..flower); or (3) with a self-controlled prolonged speech pattern (e.g., fffffflower). Interestingly, the stuttering, but not the nonstuttering speakers, showed increased right hemisphere biased activation of speech-related regions during the simulated stuttered and prolonged speech tasks, relative to the habitual speech task. In addition, between-group comparisons revealed increased right inferior frontal gyrus activation during simulated stuttering relative to nonstutterers. These data again show that we have to be very careful with linking observed neural overactivations, or laterality differences, to potential causal variables of stuttering.

Integration and Conclusion
Our research has suggested that adults who stutter, as a group, do not incorporate proprioceptive feedback as well as control participants for the control of precise movements of either oral articulation or non-oral (finger) movements. Furthermore, adults who stutter do poorer on tasks that involve learning to produce movement sequences accurately, quickly, and automatically. We have interpreted these findings as pointing towards a deficiency in integrating sensory and motor control information. This deficiency manifests itself in difficulties acquiring sequential movements, such as speech, and in developing a sufficiently high level of automaticity in their execution to allow for a fast, accurate, and fluent outcome. Brain imaging findings also suggest that adults who stutter overactivate regions of the brain involved in sensorimotor control and motor learning which is consistent with our hypothesis that speech production requires different neural resources for stuttering than fluent speaking adults. A recent report by Chang, Kenney, Loucks, and Ludlow (2009) supports the observation that such differences in neural activation are not limited to speech tasks but can be observed in nonspeech tasks as well. In addition, we have reported significantly increased activation among stuttering adult speakers in the cerebellum, known to play a critical role in integrating sensory and motor functions early on during the acquisition of motor skills (De Nil et al., 2001).

Our data also have shown that adults who stutter show a pattern of overactivation pre-treatment, even greater activation immediately following behavioural treatment, and more normalized activation patterns following maintenance treatment. Interestingly, this pattern is analogous to changes in activation patterns typically seen during early and later stages of motor learning (Rémy, Wenderoth, Lipkens, & Swinnen, 2008; Sun, Miller, Rao, & D’Esposito, 2007). This suggests that overactivation in cortical and subcortical motor areas before therapy in people who stutter could partly or wholly be a consequence of increased effort and monitoring of speech, something that stuttering individuals are known to do. For instance, Ingham, Warner, Byrd, and Cotton (2006) have observed significantly higher self-reported effort scores (and slower speech) during reading in stuttering compared to nonstuttering adults. Interestingly, these effort ratings decreased significantly for a choral reading condition, a fluency enhancing condition which also has been reported to result in more normalized brain activation patterns (Fox et al., 1996). We have hypothesized that such increased effort may be the result of an inherent deficit in stuttering speakers to acquire the necessary skill level and automaticity during speech to produce fluent speech (De Nil, 1999a). It is hypothesized that this deficit, and the resulting intermittent disruptions of fluent motor behavior, are more apparent under stressful speaking conditions, are less observable when speaking slower, and possibly result in post-treatment relapse if new speech fluency skills cannot be sufficiently automated. The deficit also may account for the fluency enhancing effects of rhythmic speech, among other fluency enhancing conditions.

Of course, many unresolved issues still remain. Not among the least is the fact that most of the data reviewed in this paper have been obtained from adults (or in a few cases, school-age children) who stutter. Whether or not preschool-age children who stutter show the same or similar responses is still very much an open question, and one that we are starting to address in our ongoing research. Also, it is unclear why the hypothesized deficiency would affect more male than female speakers. In this respect, reported gender-specific brain activation differences, such as those by Chang, et al. (2009) may start to shed some light on this issue. Finally, we have started a number of projects directly aimed at investigating the predictive value of our behavioral measures for long-term treatment outcome, and will be adding brain imaging data in the near future.

It may have been Van Riper’s intention to solve the riddle in his lifetime, but clearly it is still very much alive and undoubtedly will continue to intrigue current and future researchers and clinicians for many years. I hope that many of our young and bright students and colleagues will join us in coming years in the hunt for a solution.
Acknowledgments

The research reviewed in this paper has been supported by grants from the Natural Sciences and Engineering Research Council of Canada, the Canadian Institutes of Health Research, the Medical Research Council, and the National Fund for Scientific Research (Belgium). I would like to express my sincere gratitude to the many research participants, doctoral and masters students, and research assistants, as well as Sophie Lafaille, all of whom made this work possible.

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