Critical review

Sequence skill learning in persons who stutter: Implications for cortico-striato-thalamo-cortical dysfunction

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Abstract

The basal ganglia and cortico-striato-thalamo-cortical connections are known to play a critical role in sequence skill learning and increasing automaticity over practice. The current paper reviews four studies comparing the sequence skill learning and the transition to automaticity of persons who stutter (PWS) and fluent speakers (PNS) over practice. Studies One and Two found PWS to have poor finger tap sequencing skill and nonsense syllable sequencing skill after practice, and on retention and transfer tests relative to PNS. Studies Three and Four found PWS to be significantly less accurate and/or significantly slower after practice on dual tasks requiring concurrent sequencing and colour recognition over practice relative to PNS. Evidence of PWS’ deficits in sequence skill learning and automaticity development support the hypothesis that dysfunction in cortico-striato-thalamo-cortical connections may be one etiological component in the development and maintenance of stuttering.

Educational objectives: As a result of this activity, the reader will: (1) be able to articulate the research regarding the basal ganglia system relating to sequence skill learning; (2) be able to summarize the research on stuttering with indications of sequence skill learning deficits; and (3) be able to discuss basal ganglia mechanisms with relevance for theory of stuttering.

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1. Introduction

It has been postulated that dysfunction of the basal ganglia (BG) and its associated cortico-striato-thalamo-cortical (CSTC) connections may be an important etiological component of stuttering (Jones, White, Lawson, & Anderson, 2002; Maguire, Gottschalk, Riley, & Franklin, 2000). This proposal is based on evidence from a comparison of the symptomatology and neurophysiological characteristics reported for patients with basal ganglia dysfunction (PBG: such as patients with Parkinson’s disease, Huntington’s disease, Tourette’s syndrome, and dystonia) and persons who stutter (PWS). Some of the similarities noted between PBG and PWS include a developmental onset (Pauls, Leckman, & Cohen, 1993; Yairi, Ambrose, & Cox, 1996), environmental/situational exacerbation of symptoms (Alsobrook & Pauls, 1998; Bloodstein, 1995), facilitory effects of external sensory cues for movement (Liu, Tubbesing, Aziz, Miall, & Stein, 1999; Loucks & De Nil, 2001), facilitory effects of focused attention/de-automatization of move-
ments (Kandel, Schwartz, & Jessel, 2000; Venkatagiri, 2004), and sensitivity to fluctuating dopamine levels or dopamine-moderating medication (Kandel et al., 2000; Wu et al., 1995).

A review of brain imaging studies reveals some compelling similarities between the brain activity patterns shown by PBG and PWS. For example, a similar pattern of hypoactivation of circumscribed left motor planning areas and hyperactivation of right hemisphere homologues and bilateral cerebellar activity was found for both PBG (Berardelli, Rothwell, Thompson, & Hallet, 2001; Catalan, Ishii, Honda, Samii, & Hallet, 1999; Mentis et al., 2003) and PWS (Fox et al., 1996; Fox et al., 2000). In particular, PBG show deactivation in the left primary motor cortex and left supplementary motor cortex (SMA) (Catalan et al., 1999). PBG also show compensatory hyperactivation left prefrontal cortex, left insula, and bilateral cerebellum (Mentis et al., 2003). In addition, PBG show compensatory hyperactivation in areas responsible for attention to action including recruitment of the right hemisphere, specifically the right prefrontal cortex, SMA, and anterior cingulate cortex (Catalan et al., 1999).

PWS also consistently demonstrate hyperactivation of the cerebellum and right frontal cerebral hemisphere including the right frontal operculum (right homologue of Broca’s area), the right SMA, the right primary motor cortex, the right insula, and the right superior premotor cortex corresponding roughly to Brodmann’s area 6 (BA 6; Ingham, Fox, Costello Ingham, & Zamarripa, 2000). Some left cerebral areas are also hyperactivated by PWS including the left insula, left premotor cortex (BA 46), left prefrontal cortex, and the left anterior cingulate cortex (De Nil, Kroll, Kapur, & Houle, 2001). Activation of these areas appears to correspond to increased attention to action or de-automatization of speech movement (De Nil & Bosshardt, 2001).

Similar to PBG, PWS have also demonstrated specific deactivations, areas less active in PWS vs. fluent speakers (PNS), which have been reported in neuroimaging studies. PWS have demonstrated deactivations in Wernicke’s area, the left superior and posterior temporal lobes, and hypoactivation in circumscribed areas of the left frontal cortex including the left dorsolateral prefrontal cortices, the left superior frontal operculum (BA 44/45; Braun et al., 1997), the left inferior frontal cortex (BA 47; Fox et al., 1996; Ingham et al., 2000), the left primary motor cortex (Neumann et al., 2003, 2005; Preibisch et al., 2003), although not specifically the left SMA. Neumann et al. (2003, 2005) reported that hypoactivation of the left inferior frontal cortex and left primary motor cortex, in particular, in PWS across tasks and across pre- and post-therapy measures over a two-year period suggesting a potential locus of dysfunction.

It is well known that the neurophysiological locus of pathology in Parkinson’s disease originates in the BG complex (see Saint-Cyr, 2003 for detail). A pattern of potential loci of pathology associated with acquired stuttering was detected only recently when researchers came to consider both cortical and subcortical structures as well as their interconnections. For example, acquired stuttering cases have been reported to have resulted from damage to CSTC connections. For instance, Ludlow, Rosenberg, Salazar, Grafman, and Smutok (1987) reported that 8 of the 10 patients they examined who had acquired stuttering incurred symptoms after injury to the striatum or cortico-striatal connections. Many other authors have also reported acquired stuttering associated with damage to the BG or other structures which play a critical function in CSTC connections (Ciabarra, Elkind, Roberts, & Marshall, 2000). For example, stuttering was found to occur after damage to areas of the thalamus that project to speech control regions of the cortex (Andy & Bhatnager, 1992; Heuer, Thayer Sataloff, Mandel, & Travers, 1996; Van Borsel, Van Der Made, & Santens, 2003), the SMA (Ackermann, Hertrich, Ziegler, Bitzer, & Bien, 1996; Alexander, Naeser, & Palumbo, 1987; Van Borsel, Van Lierde, Van Cauwenberge, & Van Orshoven, 1998), or during electrical stimulation of either the thalamus or SMA (Penfield & Welch, 1951).
Additional similarities in movement characteristics have been reported for both PBG and PWS. For example, four hallmark characteristics associated with BG dysfunction include; action and rest tremor (Liu et al., 1999), involuntary movements including chorea, dyskinesia, tics, and dystonia (Abwender et al., 1998; akinesia (Low, Miller, & Vierck, 2002), and bradykinesia (Berardelli et al., 2001). Research studies indicate that many PWS also demonstrate tremor (Denny & Smith, 1992), involuntary movements (Mulligan, Anderson, Jones, Williams, & Donaldson, 2001), reaction time slowing (Smits-Bandstra, De Nil, & Rochon, 2006), and slow movement durations (Max & Yudman, 2003) relative to PNS.

In his review, Alm (2004) briefly mentioned the importance of BG systems and dopamine regulation in reward prediction during learning. What was not discussed in as much detail in his paper was the role these systems play in sequence skill learning and automaticity. Neuroimaging and primate studies have established that the BG and CSTC connections play a critical role in sequence skill learning and increasing automaticity over practice (Catalan et al., 1999). The sequence skill learning deficits of PBG are well established in the literature (Benecke, Rothwell, Dick, Day, & Marsden, 1986; Doyon et al., 1997), as are PBG’s poor performance on transfer and retention tests and on dual tasks designed to assess automaticity (Smiley-Oyen, Worringham, & Cross, 2003).

In this paper, we will first review studies that establish sequence skill learning deficits and poor automaticity development in PBG. Second, those limited skill learning and dual task studies that do exist in the stuttering literature are reviewed. Third, we will summarize the main findings from four studies specifically investigating sequence skill learning and the transition to increasing automaticity in PWS. Finally tentative conclusions surrounding the etiological implications of sequence skill learning deficits and poor automaticity development in PWS are discussed.

2. The importance of the BG for skill learning and automatization

2.1. The role of the BG in sequencing skill

A distributed system of cortical and subcortical structures is involved in sequence skill learning, retention, and transfer (see Fig. 1). Recent studies have emphasized the primary role of the BG and CSTC connections for these skills (Laforce & Doyon, 2001; see Saint-Cyr, 2003 for a review). Because evidence of BG involvement in sequence learning is well established in the literature, this paper will be limited to representative studies imparting findings most relevant to the discussion.

The basal ganglia is a complex cluster of subcortical nuclei which has extensive direct and indirect neural connections to the lateral cortex and the spinal cord (see Fig. 1). These connections have both an inhibitory and excitatory effect on neural activity. As reviewed by Alm (2004), input from the BG to the SMA, and higher order motor cortical areas via the thalamus, appears to signal the timing of practiced sequential elements (Brotchie, Iansek, & Horne, 1991; Sawaguchi & Goldman-Rakic, 1991). Research has shown that striatal neurons show rapid firing changes in the early trials of motor sequence learning. Pre-movement firing activity begins progressively earlier in these neurons during learning (Pasupathy & Miller, 2005). These striatal neurons provide a predictive signal for initiation of an increasingly automated, quick, fluent movement sequence in response to a specified stimulus (Pasupathy & Miller, 2005; Shizgal & Arvanitogiannis, 2003). Disruption of the timing of BG signaling via CSTC connections resulted in arrest, perseveration, or hesitations in fluent sequence execution in primates (Pasupathy & Miller, 2005). Weiss, Stelmach,
& Hefter (1997) found that Parkinson’s disease was associated with abnormal hesitancy and intersegmental latencies in the step-by-step performance of a sequence of hand movements. This information has led several researchers to propose that CSTC connections, particularly those of the left hemisphere, serve to integrate sensory input in order to initiate and regulate the execution timing of sequences (Roy, Saint-Cyr, Taylor, & Lang, 1993; Seitz & Roland, 1992).

Folion, Tremblay, and Bedard (1988) present a physiological explanation for disruption in the timing between BG and CSTC connections. They reported that experimentally induced dopamine loss in monkeys resulted in a remarkable loss of the specificity of movement encoding of BG neurons. When deprived of dopamine, cells in the BG lost their specificity for encoding movement direction. The firing patterns of the cells become slow, defocused, and cells responded to movements in several directions. In his review of BG function, Saint-Cyr (2003) seconded the proposal that an imbalance in dopamine regulation resulted in a delay in the cells’ ability to select appropriate responses due to ambiguity of undifferentiated choices. As seen in primates with induced lesions and PBG this defocused cell activity resulted in slow sequence initiation and hesitations/delays in sequence production during sequence learning (Pasupathy & Miller, 2005; Roy et al., 1993).
2.2. The role of the BG in early stages of learning

More recent primate studies have confirmed the importance of the BG in the earliest stages of sequence skill learning. Using single cell recording procedures, Pasupathy and Miller (2005) found that the BG were the first to encode stimulus response associations in a visuomotor learning task. They found that BG (specifically caudate) activity began earlier and earlier relative to prefrontal cortex activity during practice repetitions (a sort of anticipation) in order to facilitate quicker response initiation.

Saint-Cyr (2003) suggested that the BG are critical for finding the “ballpark,” or recognizing and selecting appropriate pre-existing movement control patterns while inhibiting irrelevant ones in early stages of learning. In support of this interpretation, Frith, Bloxham, and Carpenter (1986) had patients with Parkinson’s disease (PPD) and control subjects track a target by moving a joystick. During the first minute of each practice session control subjects showed a marked improvement in performance while PPD did not. The authors suggested that this rapid but temporary improvement in performance reflected the acquisition of a motor “set” and that PPD had difficulty in maintaining such sets.

Korman, Raz, Flash, and Karni (2003), Frith et al. (1986), and Rowe et al. (2002) concurred that CSTC connections were critical during early stage sequence learning for acquisition of set. Evarts, Shinoda, and Wise (1984) defined “set” as a matching of the parameters of a skill to suit the context in which it is being performed. Grafton, Hazeltine, and Ivry (2002) and Houk, Buckingham, and Barto (1996) proposed that the integrity of CSTC connections were critical specifically for the acquisition of set for learning complex sequential behaviours.

2.3. The role of the BG in complex/ambiguous sequence skill learning

Skill learning impairments associated with BG and CSTC connection dysfunction appear to be specific to the sequence type. Significant differences between PPD and control subjects were found predominantly for procedural skill learning. Specifically, PPD were found to have slower reaction times for implicit sequence learning tasks such as the serial reaction time tasks (SRT; see Siegert, Taylor, Weatherall, & Abernethy, 2006, for a meta-analysis). Significant differences between PPD and control subjects were also found for procedural learning tasks with a cognitive processing component, but not for declarative learning tasks (see Saint-Cyr, Taylor, & Lang, 1988 for a review).

Saint-Cyr et al. (1988) compared 24 early-stage PPD, 24 patients in various stages of Huntington’s disease, 24 healthy control subjects and 2 patients with severe declarative memory impairments. All subjects performed tests of declarative memory including parts of the Wechsler Memory Scale, the Buschke Selective Reminding Test, and a standard declarative test of verbal supraspan learning. Subjects also performed the “Tower of Toronto” test of cognitive procedural learning which requires learning of the most efficient sequence of movements to relocate four coloured discs onto three pegs (see appendix of Saint-Cyr et al., 1988). Huntington’s disease patients were somewhat impaired on both procedural and declarative tasks. However, in comparison to matched controls, PPD performed normally on all declarative tests but were specifically impaired on the Tower of Toronto. In sharp contrast, patients with declarative deficits were clearly impaired on all declarative memory tests but demonstrated no difficulties with the Tower of Toronto.

Saint-Cyr and colleagues pointed out that study methodology must attempt to control, as best as possible, the extent/stage of disease. Early stage Parkinson’s disease is thought to be
largely restricted to alteration of BG/subcortical structure function while later stages of both PPD and Huntington’s disease typically involve dementia associated with additional degeneration of cortical areas such as prefrontal areas resulting in more generalized performance deficits. In addition, similar studies comparing declarative and procedural learning skills of PPD found that the type of procedural learning task is important to detect learning deficits in PPD (Dujardin & Laurent, 2003; Harrington, Haaland, Yeo, & Marder, 1990). For example, in a study by Helmuth, Mayr, and Daum (2000) PPD and control subjects were administered a manual serial response time task. PPD were unimpaired at learning a sequence of spatial locations, but showed a deficit at learning a stimulus-to-motor-response sequence. The authors suggested that sequencing impairments in Parkinson’s disease were not general, but specific to stimulus-to-motor-response sequencing.

Further investigation also indicates significant differences between PPD and control subjects for relatively long and complex sequences involving integration of several different movements, but not for simple or repetitive sequences (Shohamy, Myers, Grossman, Sage, & Gluck, 2005). Mentis et al. (2003) compared early stage PPD with matched control subjects on a trial and error sequence-learning task. Subjects moved a mouse out to, and back from, eight spatial targets to a central location. Sequence learning was performed with timing cues so that sequence element reaction times were equivalent, however PPD demonstrated significantly more errors in the first 10 s than the control group. PPD also demonstrated a significantly slower rate of new element acquisition over the first 30 s of training relative to control subjects. These investigators also conducted positron emission tomography (PET) scanning during learning and observed that PPD demonstrated wide spread differences in brain function, specifically increased activation in preSMA, premotor cortex and insula for both right and left hemispheres relative to control subjects despite a similar number of movements in time. Mentis and colleagues suggested that these brain recruitment patterns were effective for behavioural compensation when sequences were not too long or complex and patients could take their time to learn them. They also concluded that “the dramatic failure in learning that occurs when task difficulty is high or Parkinson’s disease is advanced may occur when activation regions can no longer compensate to maintain function (p. 617)”.

Roy et al. (1993) compared PPD and age-matched control subjects on a task requiring subjects to learn a sequence of three or four different hand movements to a criterion of five consecutive correct trials. The control sequence required simple repetition of the same hand posture. The groups did not differ in their accuracy levels, however PPD demonstrated significantly slower reaction time, total movement time, and reaction time for individual movements within the sequence (inter-response time). Significant differences were found only for the different movement sequences and not for the repetitive movement sequence. Shohamy et al. (2005) found that PPD patients tested “off” L-dopa medication performed as well as matched control subjects on a first order sequence task (a task requiring learning of simple stimulus-response associations), in which subjects clicked the mouse on the correct coloured door to access the route toward a treasure. In contrast, PPD were impaired at learning the task when second order/hierarchical learning was required, where subjects clicked the mouse on a sequence of correct coloured doors to access the route toward a treasure. In contrast, PPD when tested “on” L-dopa performed equivalent to control subjects on both first and second order sequence learning.

Sequence learning deficits associated with BG dysfunction were found for speech as well as nonspeech sequencing tasks (Schulz, Leon, & Sulc, 2001; Smith & McDowall, 2004; Smith, Siegert, McDowall, & Abernethy, 2001; Westwater, McDowall, Siegert, Mossman, & Abernethy, 1998). Schulz et al. (2001) measured the accuracy and articulatory patterns of PPD relative to control subjects while learning a novel speech utterance, a nonsense phrase “Thraim po fra mo..."
dis” practiced 50 times. They found only control subjects demonstrated increased accuracy with reductions in duration and variability of productions over practice. The authors suggested that the PPD appeared to increase accuracy at the expense of reductions in movement durations and consistency. Smith and McDowall (2004) conducted a serial response time task that required verbal responses to a 12-item sequence. Twenty PPD and 37 matched controls participated. Following a visual stimulus subjects said aloud “A, B, C, or D” to indicate one of four spatial locations where the next anticipated stimulus would appear. Their results indicated that the speech reaction times of PPD improved significantly less and more gradually than those of the control subjects over practice, while the error rates for both groups were equivalent.

2.4. The role of the BG in retention and transfer

Studies with PPD have also demonstrated the importance of the BG and CSTC connections for retention (Doyon et al., 1998; Smiley-Oyen et al., 2003), and transfer (Dominey, Ventre-Dominey, Broussolle, & Jeannerod, 1997) of various skills such as finger tap sequencing or linear scaling arm movements.

Smiley-Oyen et al. (2003) compared nine PPD and matched controls on a learned linear arm movement-scaling task over two days. Subjects were required to make arm movements with their two arms proportional to the length of visually presented target bars. Scaling was acquired through knowledge of results following every second trial. PPD demonstrated poor initial acquisition of the linear scaling movement as well as significantly poorer retention of scaling skill after a retention period of 24 h. Doyon et al. (1998) compared the performance of 11 PPD and 11 matched control subjects on the serial response time task. This test consisted of a visual reaction time task with a fixed embedded sequence of finger movements. The subjects practiced the sequence 40 times per weekly session over six weeks. Eight to 16 months later subjects were retested revealing that PPD who changed from Stage 1 to Stage 2 of the disease did not show as much retention of the ability to produce the repeating visuomotor sequence. The authors determined that the difference in performance in the retention session was not due merely to a motor deterioration per se.

Based on a comparison of PPD and control subjects on a finger movement sequence learning study, Dominey et al. (1997) concluded that sequence learning and transfer appears to be impaired in PPD only when feedback/knowledge of results is not present to guide learning. They proposed that the learning mechanisms of PPD rely on augmented explicit sensory feedback. To substantiate this hypothesis, Guadagnoli, Leis, Van Gemmert, & Stelmach (2002) reported PPD required 100% knowledge of results while control subjects required only 20% knowledge of results in order to demonstrate commensurate retention of a simple timing movement.

2.5. The role of the BG in automatization

Dual-tasks are well-established in the literature as a means to investigate the level of automatization of a skill. This method assesses the level of attentional resources that a subject can allocate to a second task, as a primary task becomes well practiced and increasingly automated (Doyon et al., 1998). Thomas, Reymann, Lieury, & Allain (1996) found that PPD demonstrated poor performance relative to control subjects during early stage learning of a maze tracing and an arithmetic task, as well as after continued practice during the “automation” stage. Similarly, Doyon et al. (1997, see above) found PPD showed impairment of sequence learning after continued practice during the automatization stage. Patients with dystonia (Jahanshahi, Rowe, & Fuller, 2003), Huntington’s disease (Brown, Redondo-Verge, Chacon, Lucas, & Channon, 2001), and
PPD (Benecke et al., 1986; Doyon et al., 1998) each demonstrate poor performance on dual tasks relative to control subjects, indicating lower levels of automaticity after practice. These studies provide evidence for the continuing role of the BG and CSTC connections in the development of automaticity in sequencing skill.

3. Sequence skill learning and automatization in PWS

3.1. Sequence skill learning in PWS

Like PPD, PWS demonstrate poor skill learning relative to fluent speakers (PNS) on nonspeech sequencing tasks after practice. Weinstein, Caruso, Severing, and VerHoeve (1989) found that PWS, unlike PNS, did not show a significant decrease in the time it took to initiate the first saccade (small rapid eye movement) of a multi-saccadic sequence after practice. Webster (1986) evaluated the ability of PWS and PNS to learn 4-element finger tapping sequences and found that PWS did not show improved accuracy after practice on sequences with no repeated elements (2-1-3-4 as opposed to 2-1-3-1) compared to PNS.

PWS also demonstrate relatively slower and more inaccurate performance on speech sequencing tasks after practice relative to PNS. Ludlow, Siren, and Zikira (1997) asked PNS and PWS to practice two, 4-syllable nonsense words. Subjects practiced recalling and producing the words between trials of a listening comprehension secondary task. Ludlow et al. found that PWS were impaired in their rate of learning as well as the overall accuracy of their nonsense word productions. Similarly, Cooper and Allen (1977) found that PWS required significantly more repetitions to demonstrate increased speed of repetition for read-aloud paragraphs and sentences compared to PNS when they used between 16 and 110 trial repetitions. Children who stutter have also demonstrated a diminished ability to learn phonological sequences relative to fluent speaking children. Berman Hakim and Bernstein Ratner (2004) found that children who stutter were significantly less accurate than fluent speaking children CNS when reproducing learned nonsense syllable sequences that were three or more syllables long.

3.2. Retention in PWS

Only one unpublished study of which we are aware has compared PWS and PNS on retention of a learned motor speech task. Namasivayam and van Lieshout (2004) measured the speed and coordination of lip movements of PWS and PNS using electromagnetic articulography. PWS were asked to repeat the pronounceable nonsense words /bapi/, /bipa/, /bapiter/, and /bipater/ for 12 s with a bite block in place. When retention and transfer were assessed two and three days later, PWS demonstrated slower nonsense word performance and more variable lip coordination during production as compared to PNS.

3.3. Automatization in PWS

3.3.1. Nonspeech dual tasks

Fitzgerald, Cooke, and Greiner (1984) asked PWS and PNS to write the numbers 1–12 in a vertical line down a sheet of paper without visual feedback, simultaneously with both hands while performing a concurrent speaking task. PWS demonstrated poorly organized non-dominant handwriting, more reversed numbers, and greater differences in dominant/non-dominant hand scores compared to PNS. The authors suggested that their results could be interpreted as “the
result of an inability to allocate attention freely to processes competing for functional cerebral space” (p. 63).

Using a similar methodology, in Webster’s (1988) experiment, subjects listened to words and were asked to repeat them and then write the initial letters of the words as quickly as possible with two hands simultaneously. Results indicated that PWS made more reversed letters, showed poorer letter formation quality, and were significantly slower in responding during the dual task compared to PNS. These results were interpreted by Webster as implicating the SMA in stuttering, however he also stated that “it may be more parsimonious at this time to interpret the data in terms of group differences in processing resources” (p. 241).

Webster (1989) compared PWS’ and PNS’ ability to concurrently tap a number sequence with one hand and turn a knob back and forth 30 degrees with the other hand. PWS performed relatively poorly on both tasks under dual task conditions. Also using a tapping paradigm, Greiner, Fitzgerald, and Cooke (1986) found that PWS had slower tapping rates than PNS while simultaneously speaking. PWS demonstrated a larger dual task interference effect evidenced by increased tapping errors and increased rates of stuttered speech. In a later study Forster and Webster (2001) found PWS performed more poorly and made more on-line corrections on an “etch-a-sketch” bimanual crank task relative to PNS speakers.

3.3.2. Cognitive and linguistic dual tasks

Using a cognitive/linguistic dual task, De Nil and Bosshardt (2001) found that PWS showed significant impairment of rhyming and category decision-making during the dual task condition. PWS did not differ from PNS in the number of words used for sentence formulation, however their sentences tended to contain fewer syllables and were less grammatically complex. Neuroimaging revealed increased brain activity in areas associated with early stages of motor learning and attentive processing. These brain activity patterns suggested less automatized processing of speech/language tasks in PWS as compared to PNS.

Bosshardt (1999) asked PWS and PNS to repeat random three-word sequences containing three-syllable nouns (e.g., buttermilk) for the first task. The second task required participants to calculate the addition of three addends (22 + 12 + 3). Under dual task conditions, the stuttering rate of PWS increased beyond the base level for single task word repetition. In yet another study, Bosshardt (2002) found that the disfluencies of PWS significantly increased when simultaneously repeating words and silent reading or memorizing words.

The consistently poor performance manifested by PWS on these various dual tasks can be interpreted to suggest that PWS performed the speaking and/or the secondary tasks with relatively low levels of automaticity compared to PNS. However, this evidence speaks only indirectly to the issue. Further investigative studies which have directly evaluated the process of automatization over practice bring critical clarification to the issue and are presented below.

4. Primary objectives

Select patients with BG/CSTC connection dysfunction have consistently demonstrated impaired sequence skill acquisition and an impaired ability to develop automaticity over practice (Laforce & Doyon, 2001). If present, impaired skill acquisition in PWS may indicate possible etiological factors of stuttering. There is limited evidence in the existing literature, which speaks directly to skill learning and automaticity development in the stuttering population. To this end, four recent studies are reviewed for evidence that (1) PWS demonstrate poor speech and nonspeech sequencing skill over practice, and poor performance on retention and transfer tests compared
to PNS, and that (2) PWS demonstrate a transition to automaticity comparable to PNS during speech and nonspeech sequencing practice within a dual task paradigm. Some of these study results have been published elsewhere and details of subject recruitment and methodology can be found in the published studies (Smits-Bandstra, De Nil, & Rochon, 2006; Smits-Bandstra, De Nil, & Saint-Cyr, 2006). However, we believe that the implications of the combined study results for the involvement of BG/CSTC connection dysfunction as a possible etiological factor in stuttering are important for our understanding of the etiology and development of developmental stuttering.

5. Study One: syllable sequence reading (Smits-Bandstra & De Nil, 2006)

Twelve, English-speaking males who stutter were matched for age and compared with 12, English-speaking, male PNS. PWS scored in the very mild to moderate range (M = 16.7, S.D. = 6.8) on the Stuttering Severity Instrument-III (Riley, 1994). Participants were presented visually with a 10-syllable sequence (/ta ba pa ta ga pa ga ta pa ba/) on a computer screen and read aloud the sequence 30 consecutive times as quickly and accurately as possible. Both Practice and Group main effects were found to be significant, as well as a significant Practice × Group interaction, indicating that PNS demonstrated more improvement on syllable sequence reaction times over practice compared to PWS (see Fig. 2). Importantly, the number of errors in the 30-trial syllable sequence by PWS and PNS did not differ.

As hypothesized PWS demonstrated less improvement in speech sequencing skills after practice compared to PNS. The syllable reading reaction times of the groups were similar for the first trials but the reaction times of PWS did not improve over practice relative to PNS. This finding of PWS’ limited ability to improve speech reaction time over practice is in agreement with previous studies (Adams & Hayden, 1976; Cross & Luper, 1979; Ludlow et al., 1997). The fact that accuracy was not different for both groups, while PNS demonstrated decreased reaction time relative to PWS over practice, argues against the suggestion that differences arose from differential approaches to the speed-accuracy trade-off by the two groups (Fitts, 1954).

Fig. 2. Syllable reading sequence reaction time (ms) for PWS and PNS over 30 practice repetitions (error bars represent 1 S.D. of intersubject variability).
6. Study Two: finger-tap sequence learning (Smits-Bandstra, De Nil, & Saint-Cyr, 2006)

In this study, nine English-speaking, right-handed males who stutter were matched for age and compared with nine English-speaking, right-handed, male PNS. PWS scored in the very mild to moderate range (M = 14.8, S.D. = 7.1) on the Stuttering Severity Instrument-III (Riley, 1994). Participants were presented visually with a 10-number sequence that was randomly chosen for each individual from a pool of 3 possible sequences (/4 2 1 3 1 2 4 1 3 4/, /2 4 1 4 2 3 1 3 2 1/ or /2 1 3 2 4 1 4 2 3 1/). Subjects were instructed to use the four fingers of their right (dominant) hand to type out the numbers on four horizontally placed buttons (index finger on button 1, the middle finger on button 2, and so on). Immediately following 30 repetitions of the finger tapping sequence, subjects performed a second novel tapping sequence (the transfer sequence) 10 consecutive times. The transfer sequence was randomly selected from the remaining two sequences in the original pool. Following a 40-min retention period in which speech and reading samples were completed, participants completed the retention test. The retention test consisted of ten more trials of the original sequence. A significant quartic (4th order) Group × Practice interaction was found indicating that the finger reaction time performance curve of PNS was different than the curve of PWS (see Fig. 3).

The reaction times of PWS improved over practice but they did not approximate those of PNS. While the finger reaction times for the first trial were almost identical for both groups, PNS improved their reaction times more rapidly as compared to PWS. A non-significant trend indicated that on average, PWS did not transfer practiced improvements in reaction time to the same extent as PNS. A significant Retention × Group interaction for finger reaction time was found, indicating that PWS did not retain practiced improvements in reaction time to the same extent as PNS. The percentage errors in finger tapping across sequences by PWS and PNS did not differ significantly.

As hypothesized, PWS demonstrated poor nonspeech sequencing skill over practice, as well as poorer retention and possibly transfer skills, relative to PNS, while the accuracy rates for both groups were comparable. Relatively poor performance on retention and transfer tests by one group compared to another is commonly interpreted to suggest skill learning differences between the groups (Logan & Etherton, 1994).

![Finger Tapping Reaction Time](image)

**Fig. 3.** Finger tapping reaction time (ms) for PWS and PNS over 30 practice repetitions (error bars represent 1 S.D. of intersubject variability).
7. Study Three: dual task syllable sequence reading (Smits-Bandstra & De Nil, 2006)

Nine, native-English-speaking males who stutter were matched for age and compared with nine English-speaking male PNS. PWS scored in the very mild to moderate range (M = 17.3, S.D. = 7.6) on the SSI-III (Riley, 1994). A dual task paradigm was used in which subjects were instructed to complete two different tasks either separately or simultaneously. The two tasks used were a syllable reading sequencing task similar to Study One (see above), and a colour recognition task. The single and dual task conditions were repeated in a continuous, interleaved design (e.g., 30 trials of single task, 30 trials of dual task, 30 trials of single task, and 30 trials of dual task). The single condition syllable task was identical to Study One, with the exception that the 10-syllable ambiguous sequence was changed to /ga pa ta ba pa ta ga ba ta/. In the single task condition of the colour monitoring task, subjects were presented with a string of squares in a specific colour (e.g., red). These squares changed colours three times before disappearing from the screen (e.g., red, blue, green, red). After viewing the stimulus squares, subjects were required to indicate as quickly as they could (by saying “yes”, or “no” aloud) whether or not the same colour was presented twice.

In the dual task condition, the syllable sequence was presented in colour (e.g., red), then changed colours three times before disappearing from the screen (e.g., red, blue, green, red). The subjects read the sequence aloud as quickly and accurately as possible while at the same time paying attention to the colours. After completing the sequence, subjects were required to say aloud “yes” or “no” as quickly as they could in order to indicate whether or not the same colour was presented twice. A significant Condition x Group interaction was found indicating that the reaction times of PNS were significantly faster under single task vs. dual task conditions while the reaction times of PWS were similarly slow under both single and dual task conditions. Relatively small group differences were apparent on the first practice trial under single task conditions, before practice. However this difference between the groups grew (often more than doubled) as practice continued. No significant differences in group accuracy rates were found, however PWS demonstrated a non-significant trend toward more errors on the colour monitoring task under dual task conditions relative to PNS (see Fig. 4).

Fig. 4. Syllable sequence accuracy and colour monitoring accuracy for PWS and PNS under single and dual task conditions (error bars represent 1 S.D. of intersubject variability).
One of the most surprising and interesting findings of this study was that PWS’ reaction times, unlike those of PNS, did not differ across conditions. While PNS demonstrated two distinct performance patterns, consisting of faster reaction times over practice during the single task, and a switch back to slower reaction times during the dual task condition, the performance of PWS, in contrast, was consistently slow, and essentially similar to the dual task performance of PNS. This finding suggests that the motor planning/preparation system of the PWS was unable to transition to an increasingly efficient, relatively automated mode during task performance.

8. Study Four: finger tapping dual task (Smits-Bandstra, De Nil, & Rochon, 2006)

The twelve PNS and PWS of this study were the same subjects described in Study One (see above). Additionally, subjects were matched for typing experience (hours per day). The finger tap sequence task was identical to Study Two with the exception that the sequence used was /3 1 3 2 4 1 4 2 3 1/ printed in black. The colour recognition procedure was identical to Study Three with the exception that instead of saying “yes” or “no” aloud, subjects were required to press a “yes” or “no” button to indicate whether or not the same colour was presented twice. Two important findings of the study are presented here. A significant three-way interaction was found, which revealed that the reaction times of PNS improved more after practice compared to PWS under single task conditions only (see Fig. 5). PWS also demonstrated significantly more colour recognition errors under single and dual task conditions compared to PNS.

As hypothesized, PWS demonstrated difficulty in transitioning to increasingly automatized performance compared to PNS. PNS demonstrated steeply sloping performance curves under single task conditions and shallow, gradually sloping performance under dual task conditions. This finding suggested that PNS’ performance improvement was hampered by limited attention. PWS demonstrated a consistent, shallow performance curve across conditions. One speculative explanation for this finding was that performing even a single task for PWS required the attentional resources that would normally be reserved for dual tasks in PNS. In this study, group differences were apparent on the first practice trial under single task conditions. Similar to the results of Study

Fig. 5. Finger tapping sequence reaction time (ms) obtained by PWS and PNS over single and dual task conditions (error bars represent 1 S.D. of intersubject variability).
Three, while initial group differences were relatively small, the difference between the groups grew (often more than doubled) as practice continued.

9. General discussion

9.1. Sequence skill learning

The most significant effect found for Studies One and Two was that PWS did not demonstrate reaction time improvements over practice to the same extent as PNS. The deficits in learning efficiency were most apparent in the first 10 trials. Thus while performance was similar between the two groups on the first trial, PNS improved their performance much more quickly in subsequent trials. This early stage of skill acquisition has been termed “acquisition of set” and has been defined as a state of readiness (Evarts et al., 1984), optimization of task parameters (Friston, Frith, Passingham, Liddle, & Frackowiak, 1992), or establishing the “ballpark” of required response parameters (Saint-Cyr, 2003). Grafton et al. (2002) proposed that attention can be used to optimize adoption of set, or conversely that acquisition of set is negatively affected by lack of attention.

Acquisition of set can be observed as a steeply sloping curve of reaction time decreases over practice trials (Newell & Rosenbloom, 1981; Schmidt & Wrisberg, 2004). Venkatagiri (2004) proposed that this reaction time decrease results because movement plans/memories have been established and need only to be retrieved instead of built from ‘scratch’. This process of reaction time improvement has also been described as a result of chunking (the close binding of previously independent units; Anderson, 1982; Klapp, 2003), or strengthened/automatized stimulus-response associations (Schneider, Dumais, & Shiffrin, 1984).

Neuroscientific evidence suggests that all three of these proposals may simply be different ways of describing the same underlying phenomenon. Baddeley et al. (2000) state that memories are not inflexible structures but neurochemical traces in plastic neural networks. These traces, which develop after continued exposure to specific stimulus-response (SR) associations, become increasingly resistant to interference as they become increasingly automatized (Baddeley et al., 2000). These movement traces become increasingly efficient, or automatized, over practice.

9.2. Dual task performance

The most significant effect found for Studies Three and Four was that PWS did not demonstrate automatization of the practiced sequence to the same extent as PNS after practice. As stated earlier in this paper, neuroscientific research suggests that predictive/anticipatory signals from the BG to the frontal cortex via the thalamus facilitate the shift to increasingly automated performance over practice (Pasupathy & Miller, 2005; Shizgal & Arvanitogiannis, 2003). In accordance with this research, PNS clearly demonstrated the ability to transition to the quick accurate performance characteristic of a relatively high level of automaticity from an attention-demanding, slow, variable, and effortful performance, observed over the first 10 trials of the first single task condition. One of the most interesting findings of the current studies was that PWS, on the other hand, demonstrated only one, consistent pattern of sequencing performance regardless of condition. PWS did not demonstrate the ability to transition to quick accurate performance to the same extent as PNS. Instead, the performance of PWS was relatively slow, inaccurate, and attention-demanding as though system which triggered and facilitated the transition to increasing automaticity was impaired.
9.3. The nature of the proposed deficit

Based on a series of dual task experiments, Beilock, Carr, MacMahon, and Starkes (2002) proposed that continued conscious effort and attention focused on task performance prevents automatization (or the expression of automatization) of SR associations over practice. Neuroscientific evidence suggests that the optimization/automatization of SR binding can also be impeded when there is insufficient sensory feedback for processing of SR associations (Graybiel, 1998), or when the timing of SR associations is not consistent/predictable (Kelso, 1995). Any/all of the three explanations of poor automaticity development presented above may serve as possible explanations for the poor dual task performance and poor reaction time improvements demonstrated by PWS relative to PNS over practice.

In concordance with Beilock et al.’s (2002) proposal of conscious effort/attention impeding automatization, van Lieshout, Hulstijn, & Peters (1996) proposed that PWS choose to use a “controlled” strategy to avoid relying on an unstable motor control system. This hypothesis is based on the idea that PWS are aware that their speech system is unstable, and in reaction to this instability have developed a strategy of increased attention and effort to stabilize their speech system (Kelso, 1995). However, the current studies found PWS to demonstrate impaired performance on the finger-tapping sequencing task as well as the speech task. None of the PWS reported any awareness or concern about instability of nonspeech motor control suggesting that conscious strategy use was not likely the reason for performance differences on this task. In addition, PWS and PNS did not differ on the first practice trial when instability, anxiety levels, and the motivation to use a controlled strategy was likely the highest. Instead performance differences between the two groups grew over practice, a finding which also suggests that strategy use was not the primary reason for performance differences.

As proposed by Graybiel (1998), automatization development can be impeded by insufficient sensory feedback for processing SR associations. This second proposal is a more likely explanation for the sequence learning/automatization deficit demonstrated by PWS. This hypothesis is based on the idea that performance improvements in early stages of skill learning are thought to be reliant on accurate sensory feedback (Schmidt & Wrisberg, 2004). Early in practice movements are guided by sensory feedback. Movement is performed more slowly and the demands on attentional resources are high while sensorimotor processing is performed on-line to optimize movement parameters or “find the ballpark” (Schmidt & Wrisberg, 2004).

Both the cerebellum and the BG complex are thought to influence sensorimotor integration early in learning. Patients with cerebellar damage show poor error monitoring and self-correction during learning tasks (Fiez, Petersen, Cheney, & Raichle, 1992; Leiner, Leiner, & Dow, 1989). PPD demonstrate an incomplete automatization of the sensorimotor integration process, forcing the system to continue to guide movement by sensory input (Graybiel, 1998). Over-reliance on visual feedback is characteristic of novices, whereas experts rely predominantly on kinesthetic feedback (Beilock et al., 2002; Kelso, 1995). The automatization of sensorimotor integration builds the efficiency of kinesthetic-feedback-guided movement. The incomplete automatization of sensorimotor integration skills has been associated with impaired use of kinesthetic feedback and/or an over-reliance on visual feedback, such as is seen in the performance of novices (Smiley-Oyen et al., 2003). Both PPD (Smiley-Oyen et al., 2003) and PWS perform poorly relative to PNS on tasks requiring sensorimotor integration without visual feedback (Archibald & De Nil, 1999; De Nil & Abbs, 1991; Forster & Webster, 2001; Howell, Sackin, & Rustin, 1995; Loucks & De Nil, 2001).

As proposed by Kelso (1995), inconsistent timing of SR associations may result in decreased automatization of SR binding. This third proposal is also a likely explanation for the sequence skill
learning/automatization deficit demonstrated by PWS is that stuttering is associated with deficient optimization/automatization of SR binding due to unpredictable timing of SR associations during practice. This explanation is highly compatible with the proposal of a sensorimotor integration deficit discussed above, as the two explanations are not mutually exclusive. Instead, deficits in sensorimotor integration may result in unpredictable timing of SR associations during practice. In addition, deficits in sensorimotor integration and SR automatization (Berardelli et al., 2001; Graybiel, 1998) are both known outcomes of the dysfunction in CSTC connections associated with Parkinson’s disease.

CSTC connections are thought to play a critical role in the timing of SR associations and unit-to-unit associations in sequencing, especially in the visuomotor sequence learning tasks such as that used in the present studies. Berardelli and colleagues (2001) proposed that the role of the BG system is to reinforce and automatize SR association learning. Previous research has shown that striatal neurons show rapid firing changes in the early trials of motor sequence learning. Pre-movement firing activity begins progressively earlier in these neurons during learning (Pasupathy & Miller, 2005). These striatal neurons provide a predictive signal for increased speed of response to a specified stimulus (Shizgal & Arvanitogiannis, 2003). The interconnections between the BG complex and the frontal cortex are equally important for reinforcing associations between subsequent responses in a movement sequence as has been demonstrated by studies comparing PPD and matched control subjects on sequencing tasks (Benecke et al., 1986; Dominey et al., 1997).

9.4. Possible neurological correlates of the proposed deficit

Motor control deficits associated with PD are known to arise from deficiencies in BG function and CSTC connections. A review of neuroimaging studies of PPD reveals decreased input from the BG to the SMA and primary motor cortex via the thalamus in these patients (Berardelli et al., 2001). These brain activity patterns coincided with sequence skill learning deficits as measured by reaction time, movement duration, and accuracy (Berardelli et al., 2001). In particular, PPD demonstrated impaired acquisition of set (Frith et al., 1986).

Neuroimaging studies contrasting the brain activity patterns of PWS and PNS reveal that PWS typically demonstrate structural abnormalities (Beal, Gracco, Lafaille, De Nil, in press; Sommer, Koch, Paulus, Weiller, & Buchel 2002), aberrant timing (Walla, Mayer, Deecke, & Thurner, 2004), hypoactivation (Fox et al., 1996, 2000) of the BG (Wu, Riley, Maguire, Najafi, & Tang, 1997), and circumscribed areas of the left prefrontal and premotor cortex which form part of the CSTC connections critical for acquisition of set and sequence skill learning. Similar to PPD, the sequence learning difficulties (e.g., poor acquisition of set) evidenced by PWS may have evolved from aberrancies in these motor control structures/connections. These aberrancies disrupt the optimization/automatization of SR binding. Neumann et al. (2003) proposed that hyperactivation of the right frontal operculum specifically may compensate for deficient CSTC transmissions between Broca’s area and left-sided articular motor representations. Preibisch et al. (2003) concurred that hyperactivation of the right frontal operculum likely compensated for deficits not specific to Broca’s area but areas “downstream” from Broca’s, in particular an anomaly of white matter below the motor representation of the tongue and larynx identified by Sommer et al. (2002).

9.5. Explaining stuttering symptoms and characteristics

Any hypothesis of stuttering etiology must demonstrate the potential to further explain key defining characteristics of developmental stuttering as well as a century of literature finding
differences between PWS and PNS on tasks ranging from foot tapping to eye movements to speaking with delayed auditory feedback (Smith & Kelly, 1997).

9.5.1. Onset of developmental stuttering

Children who stutter are able to encode correctly and retrieve simple movements. They typically do not stutter when babbling, producing single words, or producing early word combinations (Bloodstein, 1995). Perhaps it is because children who stutter have difficulty retrieving sequenced movements and acquiring skill in complex sequence production that they typically start stuttering when producing words in a syntactically governed sequence (Bernstein Ratner, 1997). It is at this age when speech sequencing skills must be automatized to ensure fluent adult-like speech rate and intonation with limited susceptibility to interference from competing task demands such as language formulation.

Alm (2004) proposed that neurophysiological changes underly the ability to learn and automatize speech sequencing skills. Neuroscientific evidence suggests that learning of complex sequential skills by preschool/early school-age children likely reflects the development of the dopaminergic-dependent basal ganglia complex, CSTC connections, and the frontal/prefrontal cortex (Saint-Cyr & Taylor, 1992). Alm reviewed several studies reporting that dopamine receptor sites and receptor site availability (largely governed by the BG complex) change dramatically between the ages of 3 and 6 when children most often manifest and recover from stuttering (Seeman et al., 1987).

9.5.2. Spontaneous recovery in stuttering

Maturation of neurological structures of skill learning and automatization provides a tentative explanation not only for recovery from stuttering but also for the disproportionate spontaneous recovery of female children who stutter. The ratio of male to female adults who stutter is 4:1 while the ratio in children is approximately 1.5:1, suggesting that more girls recover from stuttering than boys (Yairi & Ambrose, 1992). Rauch and Savage’s (1997) review of anatomical studies indicated that women have larger caudate volumes than men, but this difference is the result of a gradual shift during development. Using neuroimaging, Foundas (2003) found that by age five, children have already established typically left-lateralized activation of the middle and superior temporal gyrus for auditory processing of language. It is possible that changes in lateralization and growth within the BG complex, the caudate in particular, during development may result in spontaneous recovery in some children. In support of this hypothesis are the findings of relatively decreased left-laterality of language processing areas and large caudate volumes of females in general (Rauch & Savage, 1997), and the high rate of spontaneous recovery of female children who stutter (Yairi & Ambrose, 1992).

9.5.3. Fluency enhancing conditions (FEC)

Neuroimaging studies have revealed that skill learning and complex skill production is redundantly represented in the brain with striato-thalamo-cortical circuits and cerebello-thalamo-cortical circuits (Jueptner & Weiller, 1998). These authors reported that the dominant use of the cerebellar circuit is largely determined when the skill to be learned required continuous, on-line error monitoring and reliance on sensory feedback (especially visual feedback). In contrast the BG circuit tended to be dominant for automatized tasks that are already proceduralized, internalized, and chunked into sequences. When the BG circuit is compromised, as in Parkinson’s disease, sequencing performance suffers (Benecke et al., 1986). In addition, neuroimaging studies have shown that PPD demonstrated increased activity in the
cerebellar circuit and performance reflecting dependence on the cerebellar circuit (i.e., controlled, monitored, and reliant on external sensory feedback). Alm (2004) suggested that perhaps fluency enhancing conditions such as metronomic speech, paced speech, or chorus speech reduce stuttering in PWS because they provide PWS with external sensory feedback, which cues cerebellar circuits to “kick in” and compensate for a possibly deficient BG circuit. This proposal is supported by neuroimaging results indicating hyper cerebellar activation in PWS relative to PNS (De Nil, Kroll, & Houle, 2001).

9.5.4. Treatment effects

Compensation for a dysfunctional BG circuit can also be attained by focusing conscious attention to task performance. Neuroimaging studies show that the simple act of focusing attention to motor performance and sensory feedback from motor performance significantly enhances the neural activity in the sensorimotor integration areas (e.g., the primary motor cortex, primary sensory cortex, and superior temporal gyrus for speech), and prefrontal areas in the left, and less so the right cortex during sequencing (Johansen-Berg & Matthews, 2002). This effect is also observed for PPD, and partially normalizes the decreased left frontal cortical activity in those areas due to poor input/facilitation by BG signaling.

Enhanced activity in prefrontal and sensorimotor integration areas is also seen when automatized tasks are “de-automatized” or segmented. For example Dogil et al. (2002) found that small, isolated tongue movements created more activation bilaterally in the motor and prefrontal cortices then when the tongue movements were part of an automated speech movement.

Neuroimaging of PWS reveals hyperactivation of right hemisphere SMA, primary motor cortex M1, and premotor areas (BA 6, 46), as well as hypoactivation of left inferior frontal and precentral regions and temporal regions before treatment and more normalized patterns of decreased right SMA and premotor areas and left temporal regions after treatment (De Nil, Kroll, Lafaille, & Houle, 2003). Saltuklaroglu, Kalinowski, and Guntupalli (2004) proposed that stuttering treatment and delayed auditory feedback may cause PWS to “de-automatize” or focus conscious attention to certain aspects of their speech and facilitate fluency through enhanced neural activity restoring the expected pattern of left hemisphere dominance in motor planning and sensorimotor integration areas.

9.6. Integrations of the proposed deficit with stuttering etiology theories

A deficit in the optimization/automatization of SR binding fits well with several stuttering etiology theories. The proposal that this SR binding deficit arises from the CSTC connections is an elaboration of the hypothesis of several researchers that stuttering is associated with dysfunction of BG circuits (Alm, 2004; Jones et al., 2002), including the SMA (Forster & Webster, 2001), and the dopaminergic neurotransmitter system (Maguire et al., 2000).

This proposed deficit is also highly congruent with Max’s (2004) proposal that stuttering is associated with an impaired “feedforward” mode of speech production. As reported by Weber-Fox (2001), studies comparing the electroencephalographic (EEG) records of PWS and PNS indicated that the process of stimulus perception is relatively unimpaired. Max defined stuttering as a deficit in the ability to retrieve the correct movement in response to stimuli quickly and in an increasingly automated (feedforward) manner due to an over-reliance on a slow, sensory feedback-dependent mode.

A deficit in the optimization/automatization of SR binding fits with Venkatagiri (2004), and Huinck, van Lieshout, Peters, and Hulstijn’s (2004) proposals. These authors proposed that stut-
tering is not associated with incomplete/incorrect stored movement synergies/memory traces (Venkatagiri, 2004). Rather, stuttering is a deficit in the binding/automatized retrieval of subsequent movement synergies in a sequence. For example, Huinck et al. discussed the common observation that PWS typically retrieve the initial phoneme of a word, but falter to retrieve the subsequent vowel (e.g., s-s-s-s-song). Venkatagiri cites clinical observation and reviews several studies that confirm that stuttering severity is typically reduced under conditions where a visual or auditory “priming” stimulus is present (e.g., chorus speech, orthographic text, singing).

According to the covert repair hypothesis (Kolk & Postma, 1997), stuttering results from excessive monitoring and covert prearticulatory repair of abnormally frequent speech programming errors. Neuroscientific findings are compatible with parts of the covert repair hypothesis. For example, Preibisch et al. (2003) and Ingham et al. (2000) agreed that the breakdown in sequencing within frontal and prefrontal speech control areas (areas shown to be hypoactive in neuroimaging studies) are compatible with possible deficiencies in phonological planning. Similarly, results of the current studies found delays in the reaction times of PWS also suggesting deficiencies in some aspect of output planning.

However, there is less support for Kolk and Postma’s (1997) contention that the wrong items are selected. Rather, there is evidence to suggest that PWS have delayed retrieval of syntactic and/or phonological sequence units for production which is partially overcome when primes are presented (Anderson & Conture, 2004; Wijnen & Boers, 1994).

Instead of the “excessive monitoring” hypothesis (Postma, Kolk, & Povel, 1990), the poor dual task performance of PWS in the current studies supports the demands/capacity model (Starkweather, Gottwald, & Halfond, 1990), and more specifically the ‘regression’ or ‘overload’ hypothesis of stuttering etiology (Kamhi & McOsker, 1982). These models propose that the speech skills of PWS are not stable/insufficiently automatized and stuttering results because (1) task demands exceed a limited capacity of resources, and (2) poorly automatized speech skills are susceptible to competing task demands. Bosshardt (2002) elaborated on this hypothesis, stating that the occurrence of stuttering events depended on the amount of attention required for speech and the amount of interference caused by other concurrently performed tasks that detracted attention from speech.

If stuttering is associated with a deficit in SR binding optimization/automatization, PWS would direct additional attentional resources to speech and require additional contributions from other neural systems to compensate for the deficit. Therefore, a deficit in SR binding optimization may result in stuttering depending on whether the amount of attention required for speech and any other simultaneous tasks surpasses the resource capacity. If SR binding was poorly automated, the SR associations would be relatively weak, unstable, and more susceptible to interference from ongoing activity. Poor SR binding optimization/automatization serves as a viable explanation for the large variability within and among children who stutter and PWS across time, contexts, audience, and tasks.

10. Conclusions

The present studies were designed to investigate whether or not PWS demonstrated deficits in sequence skill learning. The results from these speech and nonspeech studies present initial evidence that such deficits exist. Alm (2004) concluded that stuttering may arise from an impaired ability of the BG and/or CTSC connections to produce timing cues for the initiation of the next motor segment in speech, however the studies reviewed in this paper indicate a deficit in sequence learning and automatization, rather than a simple deficit in sequencing performance. The
neuroscientific evidence about how stimulus-response connections are developed, optimized, and automated over practice provides important information about the processes underlying sequence skill learning and dual task performance. Because the current studies were limited to observing behavioural differences between PWS and PNS, any proposals specifying underlying deficits which are potential causal factors in stuttering are speculative. Although speculative, the proposal that stuttering may be associated with a deficit in SR binding optimization and automatization serves as a viable explanation for the deficits observed in the current studies. This proposal also presents a plausible ‘common ground’ for several well-established theories of stuttering etiology.

As such, these results raise a plethora of new questions to be answered. One intriguing possibility is that the neurophysiological correlates of differences between PWS and PNS may originate in dysfunction of the distributed motor control system, more specifically CSTC connections. Based on the findings of the current studies, a direct comparison of PWS and PBG on skill learning and dual tasks is called for. Such comparisons have been supported by authors such as Ludlow and Loucks (2003) and will likely reveal important neural deficiencies and compensation that the two groups have in common. Future advances in this area will require both structural and functional brain imaging studies in order to investigate whether observed functional differences can be correlated to structural differences at the cortical and/or subcortical level, or alternatively, whether observed structural differences constitute normal intersubject anatomical variability or indeed can be associated with atypical functional activation in stuttering speakers. Furthermore, behavioural paradigms done in conjunction with neuroimaging studies with fine temporal (EEG, MEG), spatial (fMRI), and spatiotemporal measurement capabilities (e.g., electromagnetic midsagittal articulography or EMMA) are especially required to understand the complex nature of disorders of connectivity (and the resultant compensations, which stuttering is most likely to be.

CONTINUING EDUCATION

Sequence skill learning in persons who stutter: Implications for cortico-striato-thalamo-cortical dysfunction

QUESTIONS

1. Some of the similarities noted between patients with basal ganglia dysfunction (PBG) and persons who stutter (PWS) include . . .
   a. developmental onset
   b. environmental exacerbation of symptoms
   c. sensitivity to fluctuating dopamine levels
   d. all of the above
2. A review of brain imaging studies reveals some compelling similarities between the brain activity patterns shown by PBG and PWS including . . .
   a. hypoactivation of the right frontal hemisphere
   b. hypoactivation of the cerebellum
   c. hypoactivation of circumscribed left motor areas including the primary motor cortex M1
   d. none of the above
3. Similar difficulties specific to sequence skill learning reported in the literature for both PBG and PWS include:
   a. poor acquisition of set
   b. poor retention but better transfer relative to control subjects
   c. poor performance on short repetitive sequences but good performance on long complex sequences
   d. slow perception of sequential information but appropriate motor responses to sequential stimuli

4. Which of the following is a correct description of the review of the literature concerning the role of the basal ganglia (BG) and its relevant connections for sequence learning and automatization?
   a. the SMA contains tonically active neurons which signal the timing of practiced sequential elements
   b. striatal neurons provide a predictive signal for initiation of an increasingly automated, quick, fluent movement sequence
   c. the BG and its relevant interconnections serve mainly to monitor feedback and correct spatial errors of sequence movement
   d. all of the above

5. Results of recent studies conducted by Smits-Bandstra and colleagues suggest that:
   a. the speech and nonspeech sequence skill learning of PWS was impaired compared to fluent speakers
   b. PWS demonstrated decreased levels of automaticity compared to control subjects
   c. control subjects showed a sharp contrast in task performance when transitioning from low to high levels of automaticity during practice while PWS did not
   d. all of the above

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References


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