Journal of Fluency Disorders 53 (2017) 14–25

Contents lists available at ScienceDirect

Journal of Fluency Disorders

journal homepage: www.elsevier.com/locate/jfludis

Atypical non-verbal sensorimotor synchronization in adults who stutter may be modulated by auditory feedback

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ARTICLE INFO

Keywords:
Stuttering
sensorimotor control
timing
synchronization

ABSTRACT

Purpose: To investigate if non-verbal sensorimotor synchronization abilities in adult individuals who stutter (IWS) differ from non-stuttering controls (NS) under various performance conditions (tempo, auditory feedback, use of hands [single/both] and rhythm).
Methods: Participants were 11 IWS (5 males, 6 females, *Mean age* = 25.8, *SD* = 8.7) and 11 age- and gender-matched controls (*Mean age* = 24.4, *SD* = 8.4). During the experiment, participants were asked to prepare three melodies and subsequently perform them with a metronome at different rates and auditory feedback modalities (non-altered and suppressed). For each task/condition we tracked timing asynchrony related to the steady metronome beat.
Results and conclusions: : Overall, IWS displayed significantly higher timing asynchrony. Of all conditions, auditory-feedback distinguished IWS from NS most strongly, a subgroup of IWS significantly benefitting from the absence of auditory feedback. In addition, IWS showed a non-significant trend of higher negative mean asynchrony (NMA) and were more affected by the slower rate and increased rhythmic complexity and occasionally suggested poorer beat perception. These results suggest aberrant timing of sensorimotor network interaction associated with the origin of developmental stuttering.

1. Introduction

Developmental stuttering is a neurobiological disorder affecting (speech) fluency in approximately 5% of all pre-school age children and 1% of the teenage and adult population (Yairi & Ambrose, 2013). Although the exact etiology and interplay of factors in stuttering remain unclear, precise timing of movements and successful integration of sensorimotor processes appear to be necessary for effortless, fluent speech production (Civier, Tasko, & Guenther, 2010; Zelaznik, Smith, & Franz, 1994). In IWS, differences in timing and integration of sensorimotor processes have been found during both episodes of stuttering and perceptually fluent speech (Caruso, Abbs, & Gracco, 1988; Zimmerman, 1980b). There is evidence that during stutter-free speech, IWS could be identified on the basis of aberrant speech timing patterns, such as atypical rate and less accurate rhythms (Wendahl & Cole, 1961). Based on these observations, it has been proposed that impaired *timing* mechanisms may be a central component to the disorder (Andrews et al., 1983; Harrington, 1988; Kent, 1984; Van Riper, 1973). More specifically, some have argued that the core dysfunction in stuttering may be related to a defective ‘medial system’, comprised of the basal ganglia (BG) and supplementary motor area (SMA) (Goldberg, 1985), to produce ‘internal’ (i.e., in the absence of any external signal) timing cues for the initiation of the next motor segment (Alm, 2004), an hypothesis recently echoed by Etchell et al. (2014). Consequently, IWS may rely on secondary systems that utilize external

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http://dx.doi.org/10.1016/j.jfludis.2017.05.004
Received 4 November 2016; Received in revised form 13 April 2017; Accepted 19 May 2017
Available online 27 May 2017
0094-730X/

timing cues to sequence (speech) movements. Support for this viewpoint, amongst other things, comes from the fact that IWS are able to reduce their stuttering markedly when speaking synchronized to a metronome or another person – also known as 'choral reading' (e.g., Andrews, Howie, Dozsa, & Guitar, 1982). Further evidence in favor of this hypothesis has emerged from both behavioral and neuroimaging studies of speech production. For example, more temporal variability in IWS has been shown with respect to respiratory control (e.g., Zocchi et al., 1990), voice onset time (Zimmerman, 1980a), voice initiation time (Cross & Luper, 1979), vowel duration (Hand & Luper, 1980) and articulatory kinematics (e.g., Max & Gracco, 2005); these are consistent with findings of abnormal functioning of the basal-ganglia thalamo-cortical (BGTC) network in adults (Braun et al., 1997; Chang, Horwitz, Ostuni, Reynolds, & Ludlow, 2011; Watkins, Smith, Davis, & Howell, 2008) and children who stutter (Chang & Zhu, 2013), which may disappear when speaking under fluency-inducing conditions (Toyomura et al., 2011Toyomura, Fujii, & Kuriki, 2011) or after successful therapy (Giraud et al., 2008). Interestingly, areas as the BG and SMA – known to be involved, amongst other things, in the selection, initiation, timing and automatization of speech movement patterns – appear to play a role as well in rhythm and beat processing (Grahn & Brett, 2007), in line with recent findings that suggested atypical rhythm perception in children who stutter (Wieland, McAuley, Dilley, & Chang, 2015).

By contrast – but certainly not mutually exclusive with the aforementioned claims – others have posited auditory-motor coupling, sensorimotor integration and forward modeling as fundamental underlying problems of stuttering (Max, 2004; Neilson & Neilson, 1987). Consistent with this line of reasoning is the fact that IWS often immediately reduce stuttering when their own auditory feedback is altered (e.g., speaking under delayed/frequency-altered auditory feedback, masking noise or simultaneous with a second speaker; for a review, see Lincoln, Packman, & Onslow, 2006). Support for aberrant sensorimotor processes and learning in stuttering has also included findings of reduced compensation to somatosensory (Caruso et al., 1987Caruso, Gracco, & Abbs, 1987) and auditory feedback manipulations (Cai et al., 2012) as well as abnormalities in learning verbal sequences (Smits-Bandstra & De Nil, 2009), implicit speech sequence learning (Smits-Bandstra & Gracco, 2015) and overall limited retention in learning speech motor sequences (Namasivayam & van Lieshout, 2008; Smits-Bandstra & Gracco, 2015). Neuroimaging studies are generally consistent with an underlying sensorimotor abnormality by demonstrating over-activation of (primarily right-hemisphere) motor areas, increased cerebellar activity and under-activation of auditory areas (Brown, Ingham, Ingham, Laird, & Fox, 2005; Chang, Kenney, Loucks, & Ludlow, 2009; De Nil, Kroll, & Houle, 2001; Fox et al., 1996; Watkins et al., 2008), abnormal grey and white matter integrity in speech motor regions (Beal, Gracco, Brettschneider, Kroll, & De Nil, 2013; Beal, Gracco, Lafaille, & De Nil, 2007; Chang et al., 2011; Chang, Zhu, Choo, & Angstadt, 2015), atypical symmetry of the planum temporale (Foundas, Bollich, Corey, Hurley, & Heilman, 2001) and reduced functional connectivity in premotor cortical areas (Chang & Zhu, 2013; Lu et al., 2010).

One of the challenges associated with many of the aforementioned studies measuring speech motor control in IWS is dissociating whether the findings may reflect either a primary (or secondary) abnormality, or a conscious or unconscious attempt not to stutter (i.e., compensatory reaction). This problem is largely absent when one investigates performance in non-speech tasks, such as manual reaction times, finger-tapping, non-verbal dual-tasks or synchronization-continuation tasks, as IWS have generally not experienced concrete stuttering during such tasks and thus are unlikely to display any corrective or compensatory behavior.

Based on the premise that common mechanisms may be responsible for rhythmic movements across both speech and non-speech fine motor tasks (Franz, Zelaznik, & Smith, 1992), differences between IWS and controls in non-verbal timing and rhythmic tasks have been reported by some (e.g., Borden, 1983; Cooper & Allen, 1977; Falk, Müller, & Dalla Bella, 2015; Howell, Au-Yeung, & Rustin, 1997; Zelaznik, Smith, Franz, & Ho, 1997) but were not replicated by others (Hulstijn, Summers, van Lieshout, & Peters, 1992; Max & Yudman, 2003; Till, Goldsmith, & Reich, 1981). These inconclusive findings may be explained by differences in task and methodology; usually more demanding tasks elicited more pronounced differences [e.g., Hulstijn et al., 1992; Zelaznik et al., 1997] and neuroimaging may reveal differences, which may be obscured using behavioral measures.

One factor that has been overlooked in previous studies that investigated non-verbal sensorimotor behavior in IWS is the presence or manipulation of auditory feedback of self-initiated actions. Usually, tapping tasks do not elicit any complex and meaningful auditory feedback comparable to speech, raising the possibility that group differences may be modulated by this factor. Specifically, it could be that tapping or rhythmic manual movements may be less accurate and/or more variable in IWS when the complexity and meaningfulness of self-initiated auditory feedback increases. This may be a plausible possibility as both children and adults who stutter have shown aberrant auditory processing as a function of increased stimulus complexity (Corbera, Corral, Escera, & Idiazábal, 2005; Gonçalves, Andrade, & Matas, 2015). Additionally, to our knowledge, no studies to date have examined whether rhythmic complexity may be a factor that potentially affects non-verbal timing more in IWS than in controls.

1.1. Current study

Using a non-verbal synchronization task, the current experiment attempts to extend results of previous studies by investigating short-term sensorimotor learning. Here we are interested whether distinct modalities of auditory feedback (non-altered and suppressed) along with different rhythmic and manual complexity and various rates will have consequences for the timing of sequential finger movements. Participants learned to play 3 different melodies on a digital piano. The three tunes varied in complexity, with the first using only equal note values synchronized to the metronome (resembling a typical finger-tapping task), the second making alternate use of half and quarter notes and the third using both hands. This allowed us to identify also which type of complexity may result in a relative higher amount of timing asynchrony. Based on previous studies (e.g., Falk et al., 2015; Zelaznik et al., 1997), we expected that IWS would perform less synchronized to the beat than NS, especially for the more complex melodies. More specifically, if timing accuracy in stuttering is primarily modulated by rhythmic features, we expect that IWS will experience relatively more difficulty with melody 2. If specific coordination patterns (of the two hands) pose greater challenges to IWS (as shown by Zelaznik et al., 1997), we predict higher timing asynchrony scores for melody 3. Additionally, if IWS indeed show increased timing asynchrony compared to controls, we predict the former group also to exhibit a higher negative mean asynchrony (NMA), as has been shown previously by Falk et al. (2015).

Participants further played under distinct auditory feedback conditions: non-altered auditory feedback and suppressed auditory feedback (i.e., muted sound). This allows us to observe whether or not manipulations of auditory feedback may modulate ability to synchronize to an external stimulus. If stuttering comprises general (i.e., not restricted to speech) auditory-motor coupling deficiencies, we predict that the condition of suppressed auditory feedback may have a different effect on IWS than on NS, as evidenced by a large body of previous research examining the effects of altered auditory feedback on stuttering frequency (e.g., Lincoln et al., 2006).

2. Material and methods

2.1. Participants

11 IWS (5 males, 6 females, *Mean age* = 27.1, *SD* = 6.7, *range* = 19–42) and 11 age- and gender-matched controls (*Mean age* = 26.6, *SD* = 10.0, *range* = 20–46) participated in the experiment. 8 participants were of Dutch nationality and 14 of Canadian origin. None were professional musicians; however, 2 participants in each group indicated practical musical experience of +3 years. All subjects reported an absence of language, hearing and/or neurological deficits (except developmental stuttering) and current usage of drugs; all participants were right handed according to the Edinburgh handedness inventory (Oldfield, 1971) and signed for informed consent.

Reading and speech samples collected from participants in the IWS group were used to calculate stuttering severity using the Stuttering Severity Instrument-4 (SSI-4; Riley, 2009). Stuttered dysfluencies (sound and syllable repetitions, monosyllabic word repetitions, prolongations and blocks) in the samples were coded. Accordingly, 6 participants exhibited very mild stuttering severity, 1 mild stuttering symptoms, 2 moderate stuttering behaviors and 1 severe stuttering. Table 1 indicates the frequency of stuttering events (in %) and the overall stuttering severity score for each IWS.

2.2. Procedure

Three days prior to the experiment, participants received a handout containing basic instructions regarding musical parameters (rhythm and pitch), musical notation and fingering. A simplified system was used here. At the beginning of the experiment, participants were asked to confirm that they had read and understood the instructions on the handout. Possible questions were addressed and resolved before starting the experiment. As a control condition, they were also asked to complete a tapping task consisting of 30 unpaced taps at the keyboard (without sound). Participants were then provided with the sheet containing the three melodies to be performed and were told they would receive at most 15 min time to prepare each of the melodies after which each of them would be recorded in four conditions (Table 2).

Participants were also told that during the recording, a metronome on the computer was first turned on to indicate the beat and tempo to which they had to synchronize the particular melody. Participants were free to choose when to start as soon as they heard the metronome beat. When unable to continue or making a mistake (e.g., playing a wrong note or with a different finger than was intended), participants were allowed to stop and perform the melody again.

2.3. Stimuli

Stimuli were the three melodies to be performed (Fig. 1). Each melody consisted of a few bars to be played with either the right hand only (melodies 1 & 2) or with both hands (melody 3). Pitch for the first melody ranged from A to C, for the second melody from A to D and for the third melody from A to E. The first melody, containing only quarter notes, was objectively the least complex and

Table 1
Individual participant information for the stuttering group.

Stuttering participant	Gender	Age (years)	SSI score – severity	Stuttering frequency %SS
1	Female	28	25 – moderate	7.22
2	Male	29	17 – very mild	5.10
3	Female	24	19 – mild	4.56
4	Male	23	10 – very mild	3.14
5	Male	19	24 – moderate	8.52
6	Female	22	33 – severe	14.61
7	Male	32	12 – very mild	3.49
8	Female	42	8 – very mild	1.85
9	Male	33	10 – very mild	1.41
10	Female	21	8 – very mild	2.98

Note: SSI score = Stuttering Severity Instrument 4th ed. overall score. Stuttering frequency (%SS) = percent stuttered syllables averaged across the SSI speaking and reading tasks.

Table 2
Experimental conditions.

Condition	Tempo	Auditory feedback
1	Slow (IOI = 857 ms)[a]	Non-altered auditory feedback
2	Slow	Suppressed auditory feedback
3	Moderate (IOI = 666 ms)	Non-altered auditory feedback
4	Moderate	Suppressed auditory feedback

[a] IOI = inter-onset interval (in ms) between the onset of the sound of the metronome beat and that of the next one.

quasi-identical to standard finger-tapping tasks. Both the second and third melodies posed increased demands due to different levels of complexity. A (potential) difficulty in the second melody involved the unexpected half note at the beginning of the third bar. Whereas the previous two bars started each with two single quarter notes, the third bar began with a half note. The third melody required playing with two hands although this melody was rhythmically less complex than the previous one. Participants were told to be free in their manner of articulation (i.e., length of holding notes) as long as the overall rhythm remained intact. Both fingering (to which the participants had to adhere; e.g., it was not allowed to play each distinct pitch with the same finger) and names of the notes were written below and above the notes to reduce potential unnecessary levels of complexity as much as possible. Fig. 1 shows the three melodies as presented to the participants:

The control task (unpaced tapping) consisted of 30 taps by the index finger on a random key on the piano without sound. A metronome was first set at a fixed inter-onset interval (IOI) of 488 ms for 12 beats after which the participant was asked to continue tapping the same tempo.

2.4. Data analysis

All performances (including unpaced tapping) were recorded using a Yamaha digital piano, connected to the Windows program ACID Music Studio 10.0. This program translates every action on the keyboard into a number associated with the timing, pitch, loudness (key velocity) and duration. Specifically of interest was the timing as it relates to the fixed metronome beat. As the absolute duration between two consecutive beats differs depending on the tempo of the metronome beat, the program contains an additional option of using a standard scale of 0 to 767 time units between two consecutive beats (0 = on the beat). As distinct metronome rates were included in our design, we used this system for analyzing the timing asynchrony (discrepancy between the metronome stimulus and the associated key-press registration) means and standard deviations of performances of the melodies; for the control task, we calculated the mean inter-tap interval (ITI; difference between two adjacent taps) and variability of each tap in milliseconds (ms),

Fig. 1. Stimuli.
Note: A–C, …etc. refer to the notes to be played. 1–3, …etc. refers to the fingering. In the third melody, the upper staff refers to the right hand and the lower staff to the left hand.

resulting in a coefficient of variation [standard deviation/mean ITI × 100].

Of all participants' performances of the melodies, the specific timing asynchrony of individual notes were measured relative to the steady metronome beat, which differed between conditions, respectively IOI = 857 ms and IOI = 666 ms. Thus, when a participant played a note exactly on the beat, the timing asynchrony for that note would be 0. If played off the beat, asynchrony was calculated relative to the difference between the metronome stimulus and the timing of the participants' key press. For every single performance, we calculated average asynchronies for each melody and condition, which resulted in a *total timing asynchrony coefficient*, an absolute (i.e., independent of lag or anticipation of the beat) average of all asynchronies of all melodies and conditions. We additionally calculated separate total timing asynchrony coefficients, taking into consideration the direction of the synchronization (NMA).[1]

Apart from performance measurements, we also recorded how much time (in seconds) a participant used to prepare each melody. When a participant indicated a readiness to perform, the metronome beat was turned on at the appropriate rate to indicate the tempo and beat (preparatory phase). As all melodies were in the time signature of 4/4 (four beats per bar), the metronome automatically put a relative 'strong' tick on the first beat of each four. During this preparatory phase, we tracked at which bar and beat participants subsequently initiated their performance (bar/beat initiation). We further recorded how many performance attempts ('trials') were needed to achieve a performance without 'stops' or hesitations/mistakes.

As one participant in each group failed to apply the concept of playing synchronized with the metronome, we were not able to include data of these participants in our analyses. Our results were subsequently based on 10 subjects per group.

2.5. Statistical analysis

Statistical analyses were performed using IBM SPSS Statistics (version 21.0). A significance level of .05 was used. We first conducted multiple analyses of variances (MANOVA) to determine which variable differentiated the groups (IWS and NS) most strongly. We investigated 7 dependent variables:

- coefficient of variation (control task)
- timing asynchrony coefficient (averaged across three melodies),
- NMA (timing asynchrony taking into account the direction of the synchronization)
- preparation time (averaged across melodies)
- beat initiation (percentage of times initiating at a *weak* beat)
- bar initiation (percentage of times initiating *later* than 12 metronome tics of the preparatory phase[2])
- number of trials (percentage of 1st trials correct)

We then calculated the mean and standard deviations of timing asynchronies for all melodies/conditions and both groups and analyzed the results conducting 3 separate mixed repeated measures analysis of variance (ANOVA): one two-way 2 × 3 mixed ANOVA with group (IWS/NS) as the between-subjects condition and melodies (melody 1–3) as within-subjects condition and two other two-way 2 × 2 mixed repeated measures ANOVA's with group (IWS/NS) consistently as the between-subject condition and consecutively tempo (slow/moderate) and auditory feedback (non-altered/suppressed) as within subject conditions. In all mixed repeated measures ANOVA's, timing asynchrony served as the dependent variable, which displayed normal distribution and homogeneity of variance in all conditions, except for the condition of slow tempo. For IWS, we subsequently derived correlations between stuttering severity and all seven continuous variables.

3. Results

3.1. MANOVA

Table 3 displays the descriptive statistics of scores on all 7 continuous variables for both groups. The variable timing asynchrony significantly differentiated IWS from NS, $F(1,18) = 8.982$, $p = .008$, $\eta_p^2 = .333$ ($MD = 42.700$, $SE = 14.248$) at alpha levels of both .05 and .01, with a power of 0.81. In addition, there were non-significant trends ($p > .05$) of IWS more often needing a second (or third) performance attempt in order to perform a melody correctly, $F(1,18) = 3.311$, $p = .085$, $\eta_p^2 = .155$ ($MD = 9.170$, $SE = 5.039$) and IWS exhibiting a higher NMA, $F(1,18) = .932$, $p = .347$, $\eta_p^2 = .049$. There was also a tendency for a minority of IWS to occasionally initiate performance at a weak beat. It should be noted that of these 7 continuous variables, only timing asynchrony coefficient, coefficient of variation displayed normal distribution and homogeneity of variance; none of the variables correlated significantly with another ($p > .05$).

[1] When referred to (total) timing asynchrony (coefficient), we mean the asynchrony without taking its direction into consideration; otherwise we will refer to it as NMA.

[2] As the majority of the performances in both groups (90%) initiated their performance either at or during the first, second or third cycle of four beats ('bar') of the preparatory phase, initiating later was here arbitrarily considered to be aberrant.

Table 3
Descriptive Statistics on continuous variables.

	NS Mean	NS SD	IWS Mean	IWS SD
Coefficient of variation (control task)	4.54	.89	4.80	.85
Timing asynchrony coefficient*	98.30	23.92	141.00	38.18
NMA (timing asynchrony coefficient)	-20.00	30.83	-39.20	54.44
Preparation time (in sec.)	241.80	181.62	322.80	149.05
Trials (% first trials correct)	95.87	10.56	86.70	11.93
Bar initiation (% initiating later than 12 preparatory beats)	4.17	9.00	5.83	15.74
Beat initiation (% initiating on a weak beat)	0.00	0.00	5.83	10.43

* Significant at .01 alpha levels.

3.2. Mixed design analyses

Table 4 shows descriptive statistics of timing asynchronies for both groups across all conditions. They showed a clear trend of IWS displaying less timing accuracy compared to NS – i.e., IWS consistently demonstrated a tendency of higher timing asynchronies on all conditions as well as more variability (higher standard deviations). As was shown in the previous MANOVA, the two groups significantly differed on total (i.e., average across all conditions) timing asynchrony coefficients, $F(1,18) = 8.982$, $p = .008$, $\eta_p^2 = .333$ ($MD = 42.700$, $SE = 14.248$) (Fig. 2).

A two-way 2 × 3 mixed repeated measures ANOVA with group (IWS/NS) as the between-subject condition and *melodies* (melody 1, melody 2 and melody 3) as the within-subject condition showed a significant condition effect, $F(2,36) = 5.204$, $p = .010$, $\eta_p^2 = .224$ and a non-significant between group x condition interaction, $F(2,36) = 1.051$, $p = .360$, $\eta_p^2 = .055$. However, findings revealed a trend that NS appeared to experience each subsequent melody with more difficultly than the previous one, while IWS performed the least accurately in melody 2 (Fig. 3). Indeed, tests of simple effects (pairwise comparisons) using Bonferroni-correction of alpha level .017 (.05/3) revealed that IWS and NS controls only differed significantly with respect to melody 2, $p = .001$, $MD = 61.100$, $SE = 15.835$ and that of all melody comparisons, only the difference in timing asynchrony between melody 1 and 2 for IWS reached significance, $p = .001$, $MD = 47.900$, $SE = 11.596$. Individual data showed that in the NS group, 8 out of 10 people demonstrated highest asynchrony scores for melody 3, whereas in the IWS group, 6 participants performed least accurate in melody 2. Subsequent analysis showed that the interval between the first and second note of the third bar of melody 2, which had an unexpected duration of 2 counts, yielded the highest asynchrony (of all intervals) in 9 IWS; by contrast, only 5 control participants showed highest timing asynchrony at this interval.

Another two-way 2 × 2 mixed repeated measures ANOVA was conducted with group (IWS/NS) as the between-subject condition and *tempo* (slow/moderate) as the within-subject condition. There was a significant condition effect $F(1,18) = 10.599$, $p = .004$, $\eta_p^2 = .371$, suggesting that, in general, participants perceived the moderate tempo as more natural/easy to synchronize with than the slow tempo (total slow: *Mean* 134.40, *SD* 46.68; total moderate: *Mean* 104.70, *SD* 39.15). However, there was no significant between group × condition interaction, $F(1,18) = 1.154$, $p = .297$, $\eta_p^2 = .060$ (Fig. 4). Nonetheless, tests of simple effects using Bonferroni-correction of alpha level .025 (.05/2) indicated that the difference between scores for the two tempos is significant for the IWS group, $p = .007$, $MD = 39.500$, $SE = 12.902$, but non-significant for the NS group, $p = .140$, $MD = 19.900$, $SE = 12.902$, showing a stronger effect of tempo for the IWS group. Simple tests between groups showed only significance for the slow tempo, $p = .008$, $MD = 52.600$, $SE = 17.500$, demonstrating that the NS group is more accurate only in the slow tempo condition.

Finally, we performed a two-way 2 × 2 mixed repeated measures ANOVA with group (IWS/NS) as the between-subject condition and *auditory feedback* (non-altered/suppressed) as the within-subject condition. Both the condition effect and the between-group x

Table 4
Descriptive Statistics across groups and conditions.

	NS Mean	NS SD	IWS Mean	IWS SD
Melodies				
Melody 1	82.20	35.51	112.40	44.53
Melody 2	99.20	32.66	160.30	37.96
Melody 3	113.20	39.13	146.90	62.52
Auditory feedback				
Non-altered	98.40	25.26	155.50	50.35
Suppressed	97.80	27.78	126.60	35.04
Tempo				
Slow (IOI = 857)	108.10	21.03	160.70	51.19
Moderate (IOI = 666)	88.20	33.15	121.20	39.14
TOTAL	98.30	23.92	141.00	38.18

Note. Numbers refer to timing asynchrony.

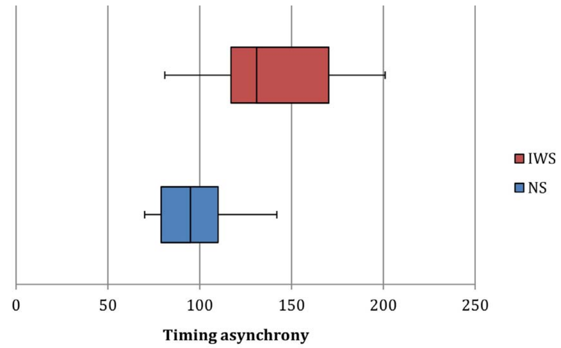

Fig. 2. Boxplot average timing asynchrony for NS and IWS.

condition interaction were (very) close to significance, respectively $F(1,18) = 4.396$, $p = .050$, $\eta_p^2 = .196$ and $F(1,18) = 4.046$, $p = .059$, $\eta_p^2 = .184$ (Fig. 5). Interestingly, whereas auditory feedback does not impact typically fluent speakers, as simple effects for NS (using alpha level of .05/2 = .025 after Bonferroni correction) showed to be non-significant, $p = .953$, $MD = .600$, $SE = 9.949$, suppression of auditory feedback may benefit at least some of those who stutter, as simple effects for IWS reached significance, $p = .009$, $MD = 28.900$, $SE = 9.949$. Inspection of individual data (Table 5) suggests that this may especially be the case for a particular subgroup within IWS; the effect of suppression of auditory feedback appears to be moderate (approximately 15-20% decrease of timing asynchrony) to robust (around 40% decrease of timing asynchrony) for 6 of the 10 subjects who stutter, whereas such considerable effects are not seen in the opposite direction (with the exception of participant 9, who performed about 29.70% *less* accurate in the suppressed auditory feedback condition). Analyses of group differences of percentages in increase/decrease with respect to auditory feedback conditions showed a close to significant larger effect in IWS than NS, $F(1,18) = 3.473$, $p = .079$, $\eta_p^2 = .162$, confirming a higher amount individual differences and within group variability in IWS. Furthermore, simple effects for between-subjects (groups) demonstrated significance only for the non-altered auditory feedback condition, $p = .004$, $MD = 57.100$, $SE = 17.569$, but not for the suppressed auditory feedback condition, $p = .053$, $MD = 28.800$, $SE = 13.896$, indicating that performance accuracy becomes closer (i.e., in this case IWS performing more accurately) when auditory feedback is suppressed, as appears to be the case with speech (Lincoln et al., 2006).

3.3. NMA

Table 6 shows descriptive statistics of NMA for both groups across all conditions. The fact that both groups exhibited negative asynchrony values for all conditions is not surprising and consistent with the sensorimotor synchronization literature (for a review, see Repp & Su, 2013). There was a non-significant trend for IWS to exhibit a higher NMA overall, $F(1,18) = .932$, $p = .347$, $\eta_p^2 = .049$. Conditions of tempo $F(1,18) = 20.370$, $p = .000$, $\eta_p^2 = .531$ displayed a significant condition effect − the slower tempo yielded a higher NMA, consistent with what is reported in the literature (Fujii et al., 2011; Repp, 2008; Zendel, Ross, & Fujioka, 2011). There was also a strikingly significant auditory feedback x group interaction, $F(1,18) = 6.509$, $p = .020$, $\eta_p^2 = .266$, where the NMA tends to increase for NS and decrease for IWS when auditory feedback is suppressed. Nonetheless, tests of simple effects did not reveal any significant results.

3.4. Correlations stuttering severity

A regression model – with stuttering severity as dependent variable and timing asynchrony coefficient, preparation time, beat

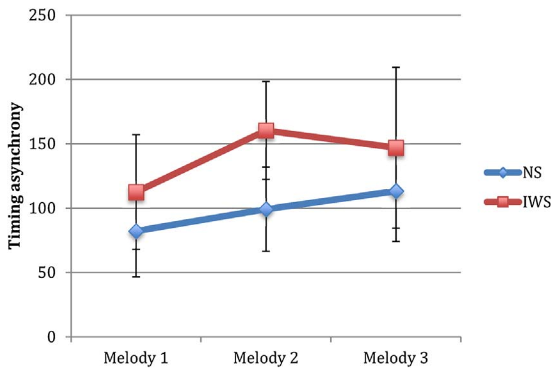

Fig. 3. Profile plot for melody condition.

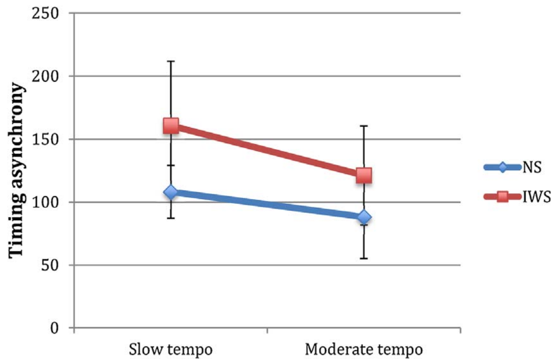

Fig. 4. Profile plot for tempo condition.

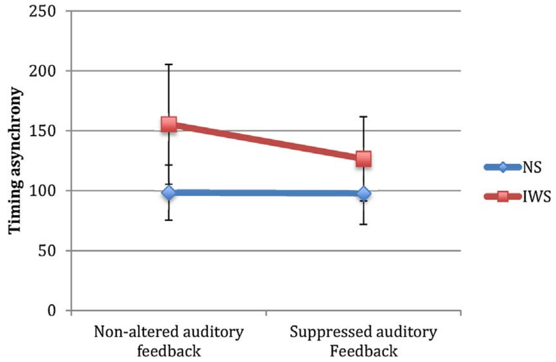

Fig. 5. Profile plot for auditory feedback condition.

Table 5
Individual data auditory feedback condition IWS.

IWS	Non-altered auditory feedback	Suppressed auditory feedback	% Increase/decrease
1	128	131	+2.34%
2	88	75	−14.77%
3	150	91	−39.33%
4	170	95	−45.71%
5	224	177	−20.98%
6	150	162	+8.00%
7	208	171	−17.79%
8	111	109	−1.80%
9	101	131	+29.70%
10	225	124	−44.88%

Note: Numbers in the second and third column refer to timing asynchrony.

Table 6
NMA descriptive statistics across group and conditions.

	NS Mean	NS SD	IWS Mean	IWS SD
Melodies				
Melody 1	−25.50	31.79	−58.70	46.24
Melody 2	−18.50	28.74	−26.90	75.25
Melody 3	−16.10	44.98	−32.30	54.25
Auditory feedback				
Non-altered	−10.50	30.48	−44.80	63.36
Suppressed	−29.60	34.53	−33.80	48.66
Tempo				
Slow (IOI = 857)	−32.50	28.36	−63.20	58.81
Moderate (IOI = 666)	−7.60	37.50	−15.40	58.34
TOTAL	−20.05	33.18	−39.30	55.27

initiation, bar initiation and number of trials as predictors (independent variables) – yielded no significant relationships, $F(6,3) = .353$, $p = .872$, suggesting no (strong) relationship between stuttering severity and the experimental variables tested here.

4. Discussion

We investigated sensorimotor synchronization abilities in IWS preparing and performing 3 melodies on a digital piano. The most important finding of this study is that IWS showed a significant higher average timing asynchrony and standard deviation compared to their fluent peers. In addition, there were clear trends of increased NMA and less timing accuracy in all conditions for IWS, with significant between-group differences for specific conditions, such as melody 2, slow tempo and non-altered auditory feedback. The results of this experiment therefore extend growing support for (generalized) timing and sensorimotor abnormalities associated with stuttering.

More specifically, of all melody comparisons, the difference in timing asynchrony between melody 1 and 2 for IWS reached significance. On average, IWS demonstrated highest timing asynchrony performing melody 2 (6 out of 10 subjects), whereas NS participants were relatively least accurate in the third melody (8 out of 10 subjects). This finding is interesting as the second melody was rhythmically the most complicated because of the unexpected half note at the first beat in bar 3. Indeed, almost all (90%) IWS subjects showed highest asynchronies at the second note of bar 3, while only 50% of the control participants did so. With respect to tempo conditions, subjects in both groups were more accurate synchronizing with a moderate metronome rate than a slow metronome rate, as we hypothesized. This was also validated by a significantly decreased NMA's for the moderate tempo compared to the slower tempo, consistent with findings from previous studies (Fujii et al., 2011; Repp, 2008; Zendel et al., 2011). Regarding timing asynchrony measures, this effect was significantly stronger in IWS, suggesting that increased deviation from a habitual rate has more impact on IWS than on controls, which parallels findings of larger group differences due to increased demands in speech (Kleinow and Smith, 2000Kleinow & Smith, 2000). Additionally, 3 IWS occasionally initiated their performance on a weak beat, whereas this was not the case with any of the control participants. As the metronome put a 'strong' tick on each first beat, starting at a weak beat (where the performance should start on the strong beat, as our melodies did not contain any upbeats) may indicate a problem with beat perception in some IWS, a possibility that warrants further investigation.

4.1. Theoretical implications

Overall, the results appear consistent with the hypothesis of stuttering as a disorder of *timing* (e.g., Etchell, Johnson, & Sowman, 2014). However, two observations may be taken into consideration when associating stuttering with a temporal abnormality. First, there was some overlap between groups on all outcome measures, except beat initiation. One of the (two) participants excluded from the analyses – due to inability to execute the task satisfactorily – was a control subject. This indicates fluent speakers without apparent morbidities may sometimes encounter serious challenges performing tapping/sequencing tasks. The variability both between and within individuals and the overlap between groups may suggest that either the underlying core mechanism may not be necessary or sufficient to develop stuttering or that the core deficiency may for some individuals indeed be speech-specific; another possibility is that those IWS whose timing asynchrony overlapped with controls perhaps may have exhibited weak or delayed motor skills in early development, but might have managed to compensate during later stages of development, yet persisting in stuttering. Stuttering is often viewed as a *dynamic* and *multifactorial* disorder (e.g., Smith & Kelly, 1997); it may be the case that for some IWS, sensorimotor skills may have normalized during adolescence and adulthood and that other factors outside the motor control domain may have been responsible for continuing stuttering behavior.

Second, as pointed out by Max (2004), various disorders with motor symptoms that do not have the temporal characteristics considered within the normal range (e.g., Parkinson's disease, idiopathic dystonia) may have distinct origins or underlying mechanisms and may therefore not be considered disorders of *timing* per se. With respect to the current task-design, Repp & Su (2013) listed that individuals with quite different disorders, such as speech/language impairment (Corriveau & Goswami, 2009), dyslexia (Thomson, Fryer, Maltby, & Goswami, 2006) and bipolar disorder (Bolbecker et al., 2011) may perform more variably in non-verbal sensorimotor synchronization tasks (i.e., tapping to a metronome). Although it was pointed out that these disorders appear to share impairments in the cerebellum as a common characteristic – which has seen evidence in stuttering as well (e.g., De Nil et al., 2001) – it remains speculative at this point whether a single particular abnormality could account for increased performance variability in those quite distinct disorders.

Our finding of an effect of suppressed auditory feedback on the performance of some IWS suggests that auditory feedback may modulate temporal accuracy to synchronize to an auditory stimulus in IWS. This finding would be consistent with theoretical models of sensorimotor dysfunction and deficient auditory-motor coupling in stuttering (Civier et al., 2010; Max, 2004; Neilson & Neilson, 1987), findings of functional and structural abnormalities in brain areas supporting auditory-motor integration in IWS (Braun et al., 1997; Chang et al., 2009; Chang & Zhu, 2013; Foundas et al., 2001; Fox et al., 1996; Watkins et al., 2008; Yang, Jia, Siok, & Tan, 2016) and the well-known fact of reduced stuttering during artificially altered auditory feedback conditions. To our knowledge, this is the first study that demonstrates a similar effect in a non-verbal sensorimotor task with IWS. Specifically, the fact that some IWS improved their performance when auditory feedback was suppressed suggests that stutterers are not overly dependent on auditory feedback, a conclusion also reached by Namasivayam et al. (2009) based on an experiment where sensory feedback of different modalities was perturbed in stutterers and non-stutterers. However, not all IWS in our study demonstrated improved performance accuracy when auditory feedback was suppressed. This appears also the case with speech, where speaking under noise and delayed auditory feedback improves fluency only to varying degrees in some persons who stutter (Andrews et al., 1982; Lincoln et al., 2006).

It may be that the hypothesized underlying mechanism may be applicable only to a sub-group of IWS. Another possibility is that the proposed deficiency may be a central core feature of stuttering but not in all IWS observable in non-verbal sensorimotor actions. Future studies may address this further by conducting concurrent experiments comparing both speech and non-speech sensorimotor behavior in not only adult IWS, but also in younger children close to the onset of stuttering, preferably over a relative longer period of time.

4.2. Limitations

One of the main limitations of this study is the small sample size. Although our main statistic of group difference with respect to timing asynchrony is associated with a power of 0.81, this result should be considered with some caution. A recent update of a study by Olander et al. (2010), where evidence for a motor timing deficit was concluded in children who stutter, failed to report a significant group difference with increased sample size (Hilger, Zelaznik, & Smith, 2016). Therefore, our results should be considered preliminary.

Ideally, our measures of non-speech synchronization ability would have been complemented with data about synchronization of spoken syllables and effects of altered auditory feedback on speech fluency. Also, inclusion of distinct modalities of altered auditory feedback (e.g., frequency and delayed) during the musical synchronization task may have given a more complete picture of the effects that altered auditory feedback may have on non-speech motor timing ability in IWS. Unfortunately, our protocol and timeframe available for this study did not allow for these additional measurements, but a potential follow-up study with increased sample size may allow addressing these issues. Further, inclusion of kinematic and electromyographic measures may provide further insight to what extent there may be significant differences between IWS and controls related to non-verbal fine motor tasks.

4.3. Conclusion

The current experiment has replicated and extended findings from previous studies (e.g., Falk et al., 2015) by reporting evidence of significant differences in sensorimotor synchronization between IWS and fluent speakers. Additionally, this is to our knowledge the first study demonstrating that in some IWS non-verbal timing asynchrony appears to be significantly modulated by auditory feedback. Overall, the results support current models of timing and sensorimotor dysfunction associated with the origin of developmental stuttering.

Conflict of interest

The authors declare that they have no conflict of interest.

Acknowledgements

We would like to thank all the participants for their time and collaboration. This work was supported by grant MOP-137001 from the Canadian Institutes of Health Research awarded to Dr. V. L. Gracco.

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