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# Adults who stutter and metronome synchronization: evidence for a nonspeech timing deficit

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**Speech timing deficits have been proposed as a causal factor in the disorder of stuttering. The question of whether individuals who stutter have deficits in nonspeech timing is one that has been revisited often, with conflicting results. Here, we uncover subtle differences in a manual metronome synchronization task that included tempo changes with adults who stutter and fluent speakers. We used sensitive circular statistics to examine both asynchrony and consistency in motor production. While both groups displayed a classic negative mean asynchrony (tapping before the beat), individuals who stutter anticipated the beat even more than their fluent peers, and their consistency was particularly affected at slow tempi. Surprisingly, individuals who stutter did not have problems with interval correction at tempo changes. We also examined the influence of music experience on synchronization behavior in both groups. While music perception and training were related to synchronization behavior in fluent participants, these correlations were not present for the stuttering group; however, one measure of stuttering severity (self-rated severity) was negatively correlated with music training. Overall, we found subtle differences in paced auditory–motor synchronization in individuals who stutter, consistent with a timing problem extending to nonspeech.**

**Keywords:** stuttering; sensorimotor; timing; speech; music

## Introduction

Stuttering is a speech disorder involving repetitions or prolongations of phonemes, syllables, or words, as well as blocks in speech. One of the characteristics of the fluent speech of individuals who stutter is a difference in speech timing evidenced by increased variability,[1–3] a lack of interarticulator coordination,[4,5] and atypical responses to perturbed auditory feedback.[6,7] Thus, it has been suggested that stuttering behavior may be related to a timing deficit that is sensorimotor in nature.[8] Coupled with this are studies showing deficits in rhythm perception[9,10] and motor sequence learning,[11] leading to the idea that this sensorimotor timing deficit may extend to nonspeech.[12] A number of studies over the years have attempted to demonstrate this by examining nonspeech timing with metronome synchronization in people who stutter. There have been equivocal results. Some report differences in interval timing, usually with those who stutter being more variable than controls.[13–18] Other work has not found any such differences.[19–22] Discrepancies between these studies have a number of possible explanations ranging from the type of populations studied to the choice of the experimental paradigm.

Olander and colleagues[16] examined synchronization behavior in a group of children (mean age = 5 years) and found greater variability in the children who stuttered. This study was followed by an update with larger sample size and slightly younger participants that found no differences between the groups.[22] However, for children around 8–9 years of age, the stuttering group and the nonstuttering group seem to diverge, with the

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stuttering group displaying more variability. Falk and colleagues[17] studied a mixture of children and adolescents (8–16 years of age) and employed circular statistics to measure synchronization differences with more sensitivity. They found that while control participants became more consistent in their synchronization as they aged, individuals who stuttered showed the opposite pattern. Intriguingly, individuals who stuttered also had a tendency to tap further ahead of the beat than control participants. Synchronizing to music seemed to attenuate the group differences. Max and Yudman[20] had earlier examined adults who stutter, reporting that rhythmic behavior was "highly similar… regardless of the presence or absence of an external pacing stimulus." However, they did not examine circular consistency, which can be more sensitive to differences in synchronization ability. They also found that the drift slopes (rate of drift from the original tempo during a phase with no auditory input) were greater for individuals who stuttered than controls, indicating that while interval variability may have been comparable, those who stutter may drift in phase. Van de Vorst and Gracco more recently studied adults and found a trend toward tapping ahead of the beat (ages 19–42), but did not manage to find significant differences in consistency.[23]

Thus, there seems to be a trend from childhood to late adolescence of individuals who stutter becoming less consistent in their synchronization than fluent speakers and also tending to respond further ahead of the beat. These trends are in need of replication in an adult population. Most of the above studies included tempi at a few different speeds, but no adaptation to tempo changes, which has been put forward as an index for sensorimotor coupling.[24] In addition, while some previous studies have measured musical training variables, the relationship of music training to synchronization in stuttering has yet to be explored.

The purpose of this paper is to evaluate the sensorimotor timing behavior of adults who stutter in order to determine whether timing problems are manifest in nonspeech behavior. Here, we include a detailed analysis of synchronization behavior in adults who do and do not stutter. We extended Max and Yudman's study by using more sensitive circular statistics, as did Falk and colleagues,[17] to examine the consistency of adults' responses and their phase relationship with the external pacing stimulus. Circular statistics treats each response as an angle relative to some zero point on a circle (zero representing the beat in this case). This frees us from the constraint of assigning each response to an audio beat and judging it as being "ahead" or "behind;" instead, we simply record its angle. Looking at the distribution of angles can lead to insights beyond the beat-to-beat patterns, implicitly taking phase drift into account. To be complete, we also measured phase drift explicitly.

It is possible that isochronous synchronization task is too simple and may not reveal differences in sensorimotor behavior between groups. In addition, synchronization behavior can vary depending on the tempo,[25,26] and group differences may only be present toward more extreme ends of the tempo range. To increase the difficulty of the task, to sample multiple tempi, and to assess the ability to recover from tempo changes, we used a paradigm where the rate of the pacing stimulus increased or decreased suddenly and unexpectedly.[24] Since synchronization abilities in general and especially mean asynchronies depend on musical training,[26,27] we made a special effort to recruit groups that were balanced for music experience, administering a music experience questionnaire and calculating the number of hours of training. We also included a short music perception test to evaluate whether music perception was similar between the groups.

## Methods

### *Participants*

Nineteen adults with a stutter (AS) and 19 adult controls (AC) were originally recruited to take part in the experiment (9 males and 10 females per group). Each participant was video-recorded as they delivered a speech sample based on the Stuttering Severity Instrument, 4th Ed. (SSI-4).[28] This consisted of some combination of reading, picture description, and spontaneous speech; at least 100 syllables were elicited for each participant (average 631 ± 227 syllables). A licensed speech-language pathologist (SLP; J.L.) conducted a blind evaluation of these videos. Five participants from the AS group had such a mild stutter that they were misclassified as AC, and four participants from the AC group displayed too many dysfluencies to be considered controls. These participants were therefore excluded. One of the remaining AS participants was identified by the SLP as having characteristics of

**Table 1. Characteristics of self-identified participants with a stutter**

| Participant | SSI-4 score | SLP classification | Self-rated severity | Self-rated anxiety |
|---|---|---|---|---|
| 1 | 23 (mild) | PWS | 5 | 3 |
| 2 | 13 (very mild) | PWS | 3.5 | 4.75 |
| 3 | 29 (moderate) | PWS | 7 | 3 |
| 4 | 17 (very mild) | PWS | 4 | 2.5 |
| 5 | 32 (severe) | PWS | 5 | 3.5 |
| 6 | 0 (none) | Control | 2 | 4 |
| 7 | 10 (very mild) | PWS | 2 | 7 |
| 8 | 13 (very mild) | PWS | 3.5 | 5 |
| 9 | 0 (none) | Control | 2.5 | 4 |
| 10 | 0 (none) | Control | 3 | 3 |
| 11 | 25 (moderate) | PWS | 4 | 4 |
| 12 | 0 (none) | Control | 4.5 | 6 |
| 13 | 8 (none) | PWS | 3.33 | 4 |
| 14 | 29 (moderate) | PWS (neurogenic) | 6.5 | 7.5 |
| 15 | 26(moderate) | PWS | 7.5 | 6 |
| 16 | 13 (very mild) | PWS | 3 | 5 |
| 17 | 8 (none) | Control | 3 | 1 |
| 18 | 14 (very mild) | PWS | 4 | 4.5 |
| 19 | 22 (mild) | PWS | 4.5 | 4.5 |

PWS, person who stutters; SSI-4, Stuttering Severity Instrument, 4th edition; SLP classification, speech-language pathologist's classification. Self-rated severity, from 1 to 9, 1 being "no stuttering" and 9 being "very severe stuttering." Self-rated anxiety, from 1 to 9, 1 being "no anxiety" and 9 being "very severe anxiety."

neurogenic stuttering and performed as an outlier on the task. In addition, an AC participant syncopated with the metronome instead of synchronizing (tapped in between beats), which, besides affecting measures of mean phase, has also been shown to be less stable and require more neural resources.[29] These two participants' data were also excluded. After exclusions, 14 AC (5 males and 9 females) and 13 AS (6 males and 7 females) were left. This is the same group of individuals reported for our recent study on vocal pitch compensation,[7] except that here one more individual is excluded because of syncopation behavior. The participants ranged in age from 18 to 51 years (mean 28 ± 10 for the stuttering group, mean 26 ± 8 for the controls; no group difference: $t(26) = -0.609$, $P = 0.548$). We report results using the smaller sample in order to be conservative and note when the larger sample differs in terms of significance. All participants were right-handed, except for one in each group.

Participants' hearing was tested in both ears, and each had hearing thresholds less than 20 dB at 125, 500, 1000, 2000, 4000, and 8000 Hz in at least one ear. They reported no neurological problems or other speech difficulties, except that one individual who stuttered had difficulty with the "r" sound during childhood (long-resolved by the time of testing). Individuals who stuttered gave a self-rating of both their stuttering severity and speaking anxiety on a scale from 1 to 9, with 1 corresponding to "no stuttering/anxiety" and 9 being "very severe stuttering/anxiety" (mean rating = 4.33 ± 1.53). Each participant responded to a questionnaire about music experience in which the number of years of music training and the average number of hours per week were given, resulting in an estimation of cumulative hours of music experience. The distribution of hours of training was left-skewed, so it was log-transformed. Both groups had similar amounts of music experience (mean of all participants = 2.64 log h, corresponding to ~437 h of experience; group comparison $t(26) = -0.699$, $P = 0.491$). Table 1 reports the characteristics of stuttering participants, including SSI-4 scores, speech-language pathology classification, self-rated severity, and self-rated anxiety.

### *Procedure*

The task involved synchronizing to a metronome sound with frequent changes in metronome rate (tempo). Participants were seated at a computer wearing Sony MDR-ZX300 over-the-ear headphones.

In each trial, participants were presented with a binaural metronome stimulus consisting of 12 different tempi, with 10–14 clicks per tempo (the exact number of clicks was determined at random for each tempo). Click volume was adjusted for the participant's comfort, ranging between approximately 67 and 76 dB, though certain subjects went as low as 55 dB (values were measured with an A-weighted sound-level meter using the fast response setting). The interbeat interval (IBI) for each tempo ranged from 350 to 700 ms in steps of 25 ms (a range of approximately 86–171 beats per minute). These values span much of the range of tempi used in music and include suggested values for the average human's spontaneous or preferred tempo.[30–32] Tempo changes were constrained so that in each trial, an equal number of speed-up and slow-down changes were present, and that each of several magnitudes of tempo change was equally represented (75, 100, 125, and 150 ms). This resulted in no tempo changes being too drastic; for example, the tempo never changed from a 400-ms interval to a 700-ms interval directly. Participants completed five or six trials, resulting in a total of 60–72 different tempi presented, and 55–66 instances of tempo change.

Participants responded to the metronome stimuli by squeezing a small pressure pad held between the index finger and thumb (Biopac® precision transducer, parts TSD160A & DA100C). Using a pressure pad ensured that there was no auditory feedback from the subject's own production. Individuals who stutter respond differently to auditory feedback during synchronization,[23] but in the current study, we were interested in the sensorimotor ability of individuals who stutter to generate synchronized motor output in response to an external auditory cue, rather than assessing auditory feedback. The pressure pad tracked the changes in pressure rather than the absolute pressure. Using a splitter, the auditory signal from the headphones was fed into an audio interface, along with the simultaneous signal from the pressure pad, so that they could be aligned with each other in time. The synchronization task lasted about 15 minutes.

Finally, participants completed the Montreal Battery for the Evaluation of Musical Abilities (MBEMA), a short test for basic musical perception.[33] This included subtests for melody, rhythm, and memory. For the melody and rhythm subtests, participants listened to two melodies and were asked if they were the same or different. The differences were either changes in note pitch for the melody section or changes in note rhythm for the rhythm section. For the memory subtest, participants were presented with a melody and were asked if they had heard it in either of the previous sections.

### *Analyses*

Analyses were performed using MATLAB® R2015b (MathWorks®, Inc., 2015).[34] In a series of preliminary tests, it was determined that the tubing connecting the pressure pad to the Biopac caused a constant delay in the response signal of 37.5 ms; this delay was subtracted out before analysis. Discrete peaks of the response signal (blue trace in Fig. 1A) were extracted using the *findpeaks* function in MATLAB. We set the minimum threshold for the peak prominence to the root mean square (RMS) value of the entire trial (RMS is a measure of energy in the signal). The Biopac system did not measure absolute pressure, but rather its first derivative, so the signal represents pressure change. Using the peaks of the signal resulted in asynchronies that were similar to previous tapping literature.[26]

From this set of responses, the *interresponse interval* (IRI, the distance from one response to another; see Fig. 1B) was measured, as well as *phase angle* (measuring the relative distance of each response from the closest audio beat; see Fig. 1C). IRI was analyzed linearly, but phase measures were analyzed using the circular statistics toolbox in MATLAB.[35]

**IRI accuracy.** For all analyses at a steady tempo, we used the values of all responses except the first two following each tempo change (for more information on why we excluded the first two responses, see Figs. S1 and S2, online only). To calculate the IRI error, the following equation was used:

$$\% \text{ IRI error} = (|\text{IRI} - \text{IBI}|\,/\text{IBI}) \times 100,$$

where IRI is the interresponse interval, and IBI is the corresponding interbeat interval from the audio signal, both measured in milliseconds.

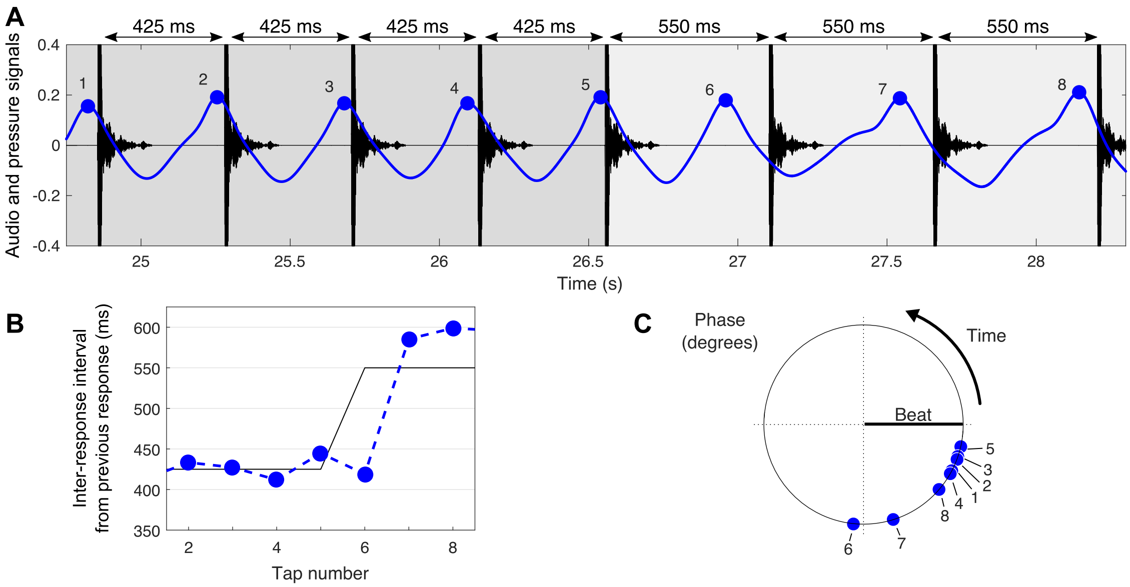


**Figure 1.** Sample portion of a metronome stimulus and response, including a tempo change. (A) Over the metronome audio (black) is plotted the response data (blue), representing a change in pressure on the pressure pad over time. Peaks are indicated by circles. (B) The interresponse interval (IRI) is calculated between each peak and the peak that precedes it. (C) Phase angles (i.e., asynchronies) of the responses compared with the metronome stimulus, expressed in degrees. The tempo change on beats 6 and 7 causes a delayed correction of IRI in (B) and a temporary angular displacement in (C).

**Phase angle and phase consistency.** The phase of each response was determined by the following:

$$\text{Phase angle} = (\text{asynchrony}/\text{IBI}) \times 360 \text{ degrees},$$

where asynchrony (difference in time relative to the closest beat) and IBI (interbeat interval) are both measured in ms, and asynchrony can be positive or negative depending on its relationship to the closest beat. Using this measure made it possible to compare asynchrony values across different tempi and measure ambiguous responses without worrying about which audio beat they were assigned.

A mean resultant vector was calculated by treating each phase measurement as a unit vector, adding all of the vectors together, and dividing the length of the resulting vector by the number of measurements:

$$\text{MRL} = (1/N) * \Sigma r_i,$$

where $r_i$ is a single response vector and $N$ is the number of responses. The angle of this mean resultant vector was the *mean phase angle*, and the mean resultant length (MRL) described the *mean phase consistency* on a scale from 0 (no consistency) to 1 (perfect consistency). Because of the hard boundaries at 0 and 1, MRL may have compressed variance at its extreme ends, similar to proportional data. Therefore, we transformed the data using the logit function before performing statistical analyses (this did not change the significance of any results compared with using the raw values). The more intuitive raw value for MRL is shown in figures.

Mean %IRI error was calculated for each participant, along with mean phase angle and mean phase consistency. To look at steady-state synchronization behavior at different tempi, we established three tempo categories, evenly dividing the presented tempi into fast (350—450 ms intervals), medium (475—575 ms), and slow (600—700 ms). Percent IRI error, mean phase angle, and phase consistency were averaged per participant for each tempo type (circular averages were used for circular measures). A two-way ANOVA or a high-kappa test (the circular form of a two-way ANOVA) was used to test for main effects of group and tempo, as well as the interaction.

**Phase drift.** Phase drift (i.e., progressive change in phase angle) was assessed using the slope of a best-fit line through the 3rd to 10th responses following each tempo change. A two-way ANOVA was performed on the absolute value of these slopes

**Table 2. Results of ANOVAs on variables of interest**

| Measure | Mauchly's test of sphericity | Main effect of group | Main effect of tempo | Interaction |
|---|---|---|---|---|
| IRI accuracy | $\chi^2(2) = 7.2, P = 0.027$ | **$F(1,25) = 3.2, P = 0.085, \eta^2_G = 0.10$** | $F(1.6,39.7) = 2.5, P = 0.106, \eta^2_G = 0.01$ | $F(1.6,39.7) = 1.8, P = 0.184, \eta^2_G = 0.01$ |
| Mean phase angle | NA (circular data) | **$F(1,25) = 5.9, P = 0.023$** | **$F(2,50) = 8.0, P = 0.001$** | $F(2,50) = 2.0, P = 0.152$ |
| Phase consistency (MRL) | $\chi^2(2) = 6.8, P = 0.033$ | $F(1,25) = 2.0, P = 0.168, \eta^2_G = 0.06$ | $F(1.6, 40.1) = 0.1, P = 0.838, \eta^2_G < 0.01$ | **$F(1.6, 40.1) = 6.1, P = 0.008, \eta^2_G = 0.04$** |
| Phase drift | $\chi^2(2) = 5.4, P = 0.068$ | $F(1,25) = 0.4, P = 0.532, \eta^2_G = 0.01$ | $F(2,50) = 0.7, P = 0.495, \eta^2_G < 0.01$ | $F(2,50) = 0.9, P = 0.425, \eta^2_G < 0.01$ |

NOTE: Effect size is given as generalized eta squared. If Mauchly's test of sphericity is significant, subsequent statistics are corrected. Since mean phase angle is a circular measure, neither Mauchly's test nor generalized eta squared is available. Cells with significant results or trends are bolded.

to examine the effects of group and tempo. For phase drift analyses, angles on individual beats were allowed to continue past ± 180 degrees if doing so allowed them to be closer to the preceding angle, using the *unwrap* function in MATLAB. In addition, we visually inspected the phase behavior of individual participants to identify different phase-drift behaviors.

**Tempo changes.** At tempo changes, an overshoot response was measured. Overshoot behavior consisted of producing intervals more extreme than the actual change in tempo; for example, if the intervals became slightly larger (slower tempo), a participant would produce intervals even larger than the new tempo for the first few beats, before relaxing into the new tempo. This is usually necessary to make up for an incorrect interval produced at the moment of the tempo change and recover one's phase angle; in fact, the difference between the first and second beats after a tempo change is usually taken as an indicator of the strength of sensorimotor coupling.[24] Here, the overshoot was measured in terms of IRI change relative to the previous tempo during the first three responses after the new tempo with an ANOVA: 2 [group] × 3 [tap] × 2 [speed up versus slow down], with interactions.

### *Correlation analyses*

Correlations between steady-state measures (%IRI error, phase angle, phase consistency) and between these measures and music measures (hours of experience, perception score) were also calculated. For correlational analyses of phase angle with other linear measures, we took the cosine of each participant's angle to represent how far away the response was from the beat while avoiding 180-degree differences that could compromise the correlation.

All *t*-tests are two-tailed. The effect size for ANOVAs was measured using generalized eta squared ($\eta^2_G$),[36,37] with the exception of phase angle, which is a circular measure and for which we are not aware of an appropriate effect size measure. For phase angle, we have instead reported the means for both groups in the text.

## Results

ANOVA statistics on IRI accuracy, mean angle, consistency, and drift are presented in Table 2.

### *IRI accuracy*

AS had a mean error of about 6.0% of the tempo versus 4.9% for AC, indicating less accurate reproduction of intervals for the AS group on average (see Fig. 2B, top panel). There was a trend toward the main effect of group, a trend for an effect of tempo, and no interaction. Note that the group difference was not significant with the larger sample ($P = 0.253$), but the effect of tempo became significant ($P = 0.008$).

### *Mean phase angle*

Individuals who stuttered had a mean phase angle of −76.8 degrees, much more negative than AC (−38.0 degrees). As seen in Figure 2A and B (middle panel), there was the main effect of group and the main effect of tempo, but no interaction. Both groups were closer to the beat (less negative mean asynchrony) at fast tempi, though again AS exhibited a more negative mean phase angle than AC. This difference existed despite considerable individual variability. Figure 2A shows the

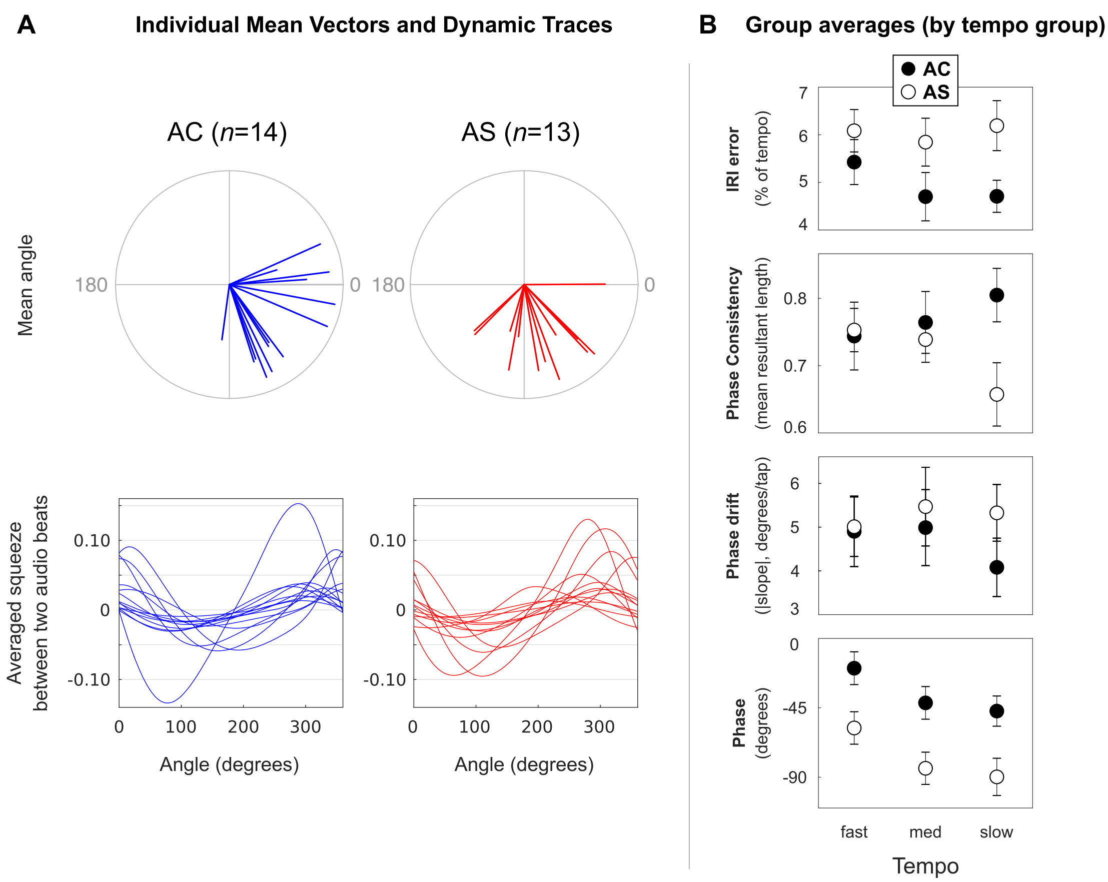


**Figure 2.** (A) Mean resultant vectors for each participant for stable portions of tempo, showing both mean phase angle and phase consistency (indicated by the length of each vector). Individuals who stutter (red) have significantly more negative phase angles relative to controls (blue). Below the circular plots are the averaged dynamic traces for each participant between audio beats, demonstrating consistency with the peak-finding analysis. (B) Average IRI percent error, phase consistency, phase drift, and phase angle by tempo category. Black circles: control participants; empty circles: stuttering participants (error bars indicate the standard error). AS display a trend toward larger IRI errors than AC, and are more negative in their mean phase angle (i.e., they respond earlier than AC on average). AS are also less consistent than AC at slow tempi in particular.

angular asynchronies of individual participants (for all tempo categories put together), along with the average signal trace per subject, which shows that the peak-finding and signal-averaging methods give similar results for phase angle.

### *Phase consistency*

There was no main effect of group and no main effect of tempo, but there was a significant interaction between tempo and group. Post hoc tests showed no effect of group at fast tempi ($F(1,25) = 0.1$, $P = 0.747$), and no effect of group at medium tempi ($F(1,25) = 0.8$, $P = 0.370$), but there was a significant effect of group at slow tempi ($F(1,25) = 6.3$, $P = 0.019$). In addition, the simple effect of tempo was not significant for AC ($F(2,24) = 1.9$, $P = 0.174$), but it trended toward significance for AS ($F(2,24) = 2.6$, $P = 0.097$). In summary, AS were more affected by tempo, such that they became more inconsistent in their angle than AC at slow tempi. Again, individual variability can be seen in Figure 2A by observing the varied lengths of the vectors. Note that the post hoc difference between groups at slow tempi just missed significance with the larger sample ($P = 0.066$).

### *Phase drift*

Based on our observations from Max and Yudman's study,[20] we examined the degree to which the participants drifted in phase during the steady-state portion, still excluding the first two taps after tempo change. The results are illustrated in Figure 2. There was no main effect of group, no effect of tempo, and no interaction.

Upon examining results for individual subjects, we did observe a few different behavioral patterns in the steady-state portion following the tempo change (Fig. S4, online only). Some individuals demonstrated what we call "phase attraction;" they seemed to return to a preferred phase angle after a tempo change and continued to match that phase angle even more tightly over the course of the steady-state portion. A "phase drifter," on the other hand, corrected somewhat for phase at the tempo change but gradually drifted in phase angle during the steady-state portion. Finally, there is the curious case of "phase-flexible" behavior, in which the participant seemed to take on and maintain a new phase after each tempo change, based on the effect of the shortening or lengthening interval. There was not a striking difference in the number of individuals fitting each type, but this could merit some further study with a larger group of participants.

### Tempo changes

An overshoot effect was observed in response to tempo change, as in Repp's seminal work,[24] such that participants first overcorrected their IRI in order to make up for any discrepancy, and then returned to the correct IRI after a few beats, a pattern remarkably similar for both groups (Fig. 3A, top panel). There was a difference in the pattern for speeding up versus slowing down (Fig. 3A, bottom panel). When the tempo slowed down (larger intervals; positive-going curves in Fig. 3A), the overshoot usually peaked at the first response after the tempo change. However, when the tempo sped up (smaller intervals, negative-going curves in Fig. 3A), the overshoot usually peaked on the second response after the tempo change.

A three-way ANOVA (group, response after change, and change direction) was performed on the overshoot data (Fig. 3A, bottom panel). Mauchly's tests of sphericity were significant for the main effect of response ($\chi^2(2) = 25.0$, $P < 0.001$), and the interaction of response and change direction ($\chi^2(2) = 6.9$, $P = 0.032$), so a Greenhouse–Geisser correction was applied. There was no main effect of group ($F(1,25) = 0.2$, $P = 0.679$, $\eta_G^2 < 0.01$) and no interactions between group and any other factors ($P \geq 0.440$). There was a main effect of response ($F(1.2,30.0) = 12.7$, $P = 0.001$, $\eta_G^2 = 0.18$), which strongly interacted with change direction ($F(1.6,40.0) = 29.7$, $P < 0.001$, $\eta_G^2 = 0.18$), reflecting the differential delay in correction between speed-up and slow-down tempo changes. The three-way interaction was not significant ($F(1.6,40.0) = 0.6$, $P = 0.532$, $\eta_G^2 < 0.01$).

### Correlations between steady-state behaviors, music measures, and severity

Table 3 shows the correlations between the different measures extracted from the steady-state portion (for correlations from the larger sample, see Fig. S3 and Table S1, online only). IRI was associated with MRL, music training hours, and music perception scores (MBEMA total score) for the control participants only.

Stuttering severity (self-rated) correlated with phase angle, though for phase angle the correlation was in the opposite direction than one would expect given the group difference. In other words, one might expect that individuals with the most severe stutter would have the most negative phase angle (since AS as a group had more negative phase angles than AC). However, we observed the opposite: those with the most severe stutter had the least negative phase angle. We also looked at the correlation between phase consistency at slow tempi only and stuttering severity in AS (where we had observed a group difference): it was not significantly correlated with either self- or SLP-rated severity ($P \geq 0.585$). Interestingly, stuttering severity was also correlated with music training, but not at all with music perception.

### Music perception

A 2 × 2 ANOVA on the MBEMA test (group, subtest with levels melody/rhythm/memory) was performed. Mauchly's test of sphericity was not significant for the effect of subtest ($\chi^2(2) = 1.9$, $P = 0.393$). There was a trend for an effect of group ($F(1,25) = 3.4$, $P = 0.079$, $\eta_G^2 = 0.06$), with individuals who stuttered scoring slightly lower across the board (AC average score: 18.4 ± 1.1; AS average score: 17.4 ± 1.7). The main effect of subtest ($F(2,50) = 1.2$, $P = 0.309$, $\eta_G^2 = 0.02$) and the interaction ($F(2,50) < 0.1$, $P = 0.982$, $\eta_G^2 < 0.01$) were

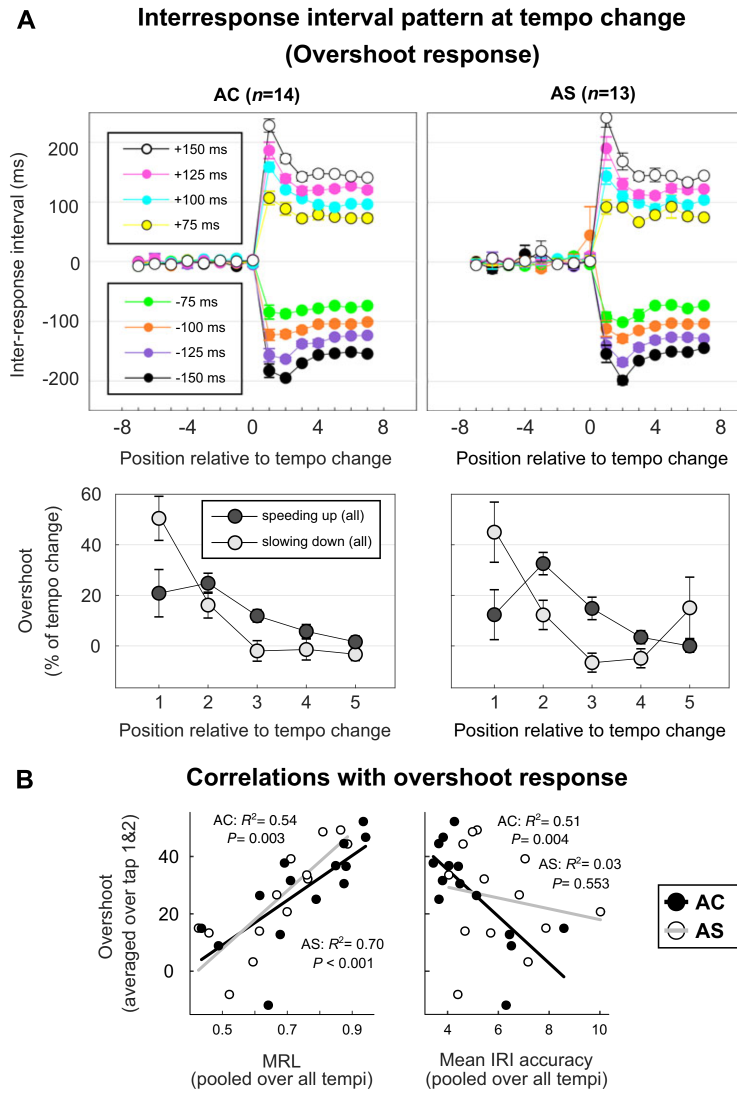


**Figure 3.** (A) Overshoot dynamics at tempo change. These graphs plot average IRI relative to the IBI of the previous tempo ((IRI – IBIprev)/IBIprev). Each magnitude of tempo change is represented separately in the top panel. In the bottom panel, IRI is shown as a percentage of the tempo change for all accelerations (black) and decelerations (gray). For example, a 40% overshoot for a tempo change of +100 ms would mean that, on average, subjects produced an interval 140 ms longer than the previous tempo, rather than 100 ms longer. Negative changes in tempo (i.e., speeding up) are multiplied by –1 in this panel so they can be compared with positive changes in tempo (slowing down). (B) Correlations of the overshoot response with angular consistency (MRL) and mean IRI accuracy. Overshoot is tightly correlated with MRL regardless of group. Overshoot is tied to IRI accuracy for AC but not for AS.

not significant (note: these results not shown in a figure). Performance on the MBEMA was at ceiling in many cases, which may have obscured some differences in music perception. Note that the group difference became significant in the larger sample ($P = 0.048$).

## Discussion

The idea that stuttering is a disorder of timing is not new.[12] The problem is that abnormal *production* in nonspeech timing tasks (e.g., to a metronome) has proven difficult to consistently demonstrate in

**Table 3. Correlations between measures for each group**

| | Phase (cos of angle) | | IRI error | | Music training | | MBEMA | | Severity (SLP) | Severity (self) |
|---|---|---|---|---|---|---|---|---|---|---|
| **Group** | AC | AS | AC | AS | AC | AS | AC | AS | AS | AS |
| MRL | $R = 0.38$<br>$P = 0.185$ | $R = 0.46$<br>$P = 0.111$ | $\mathbf{R = -0.78}$<br>$\mathbf{P = 0.001}$ | $R = -0.28$<br>$P = 0.353$ | $R = 0.49$<br>$P = 0.073$ | $R = -0.05$<br>$P = 0.864$ | $R = 0.52$<br>$P = 0.059$ | $R = -0.10$<br>$P = 0.734$ | $R = 0.23$<br>$P = 0.452$ | $R = 0.16$<br>$P = 0.605$ |
| Phase (cos of ∠) | | | $R = 0.06$<br>$P = 0.843$ | $R = -0.01$<br>$P = 0.973$ | $R = 0.11$<br>$P = 0.700$ | $R = -0.36$<br>$P = 0.225$ | $R = -0.23$<br>$P = 0.423$ | $R = -0.20$<br>$P = 0.515$ | $R = 0.53$<br>$P = 0.060$ | $\mathbf{R = 0.60}$<br>$\mathbf{P = 0.032}$ |
| IRI error | | | | | $\mathbf{R = -0.55}$<br>$\mathbf{P = 0.039}$ | $R = -0.23$<br>$P = 0.450$ | $\mathbf{R = -0.74}$<br>$\mathbf{P = 0.003}$ | $R = -0.35$<br>$P = 0.243$ | $R = -0.02$<br>$P = 0.957$ | $R = 0.08$<br>$P = 0.793$ |
| Music training | | | | | | | $R = 0.22$<br>$P = 0.444$ | $R = 0.02$<br>$P = 0.946$ | $R = -0.38$<br>$P = 0.199$ | $\mathbf{R = -0.71}$<br>$\mathbf{P = 0.006}$ |
| MBEMA | | | | | | | | | $R = -0.02$<br>$P = 0.959$ | $R = 0.01$<br>$P = 0.982$ |
| Severity (SLP) | | | | | | | | | | $\mathbf{R = 0.78}$<br>$\mathbf{P = 0.002}$ |

NOTE: White cells indicate correlations for control participants, and gray cells indicate individuals who stutter. Bolded text in cells indicates significant correlations at $P < 0.05$ uncorrected. Bonferroni correction: $0.05/31 = 0.0016$ (with this correction, only the association between MRL and IRI in AC is significant, though IRI/MBEMA for AC and music training/self-rated severity for AS come close).

AS, adults with a stutter; AC, adult controls; MRL, mean resultant length (i.e., phase consistency); IRI, interresponse interval; MBEMA, music perception test; SLP, speech-language pathologist.

this population. In the current study, we found IRI errors that were very low, around 5%, regardless of tempo, with the IRI production ability of individuals who stutter being only slightly more variable than that of control participants. However, taking phase into account, we saw additional group differences. Individuals who stutter tended to anticipate the beat more than fluent adults, confirming trends in recent work.[17,23] Moreover, by using a circular measure (phase consistency, or MRL) that simultaneously evaluates interval and phase angle, we found that AS synchronization diverged from AC synchronization the most at slower tempi. Individual patterns of drifting or flexible phase behavior (Fig. S4, online only) along with a marginal increase in IRI variability (Fig. 2) might help to account for this lower consistency in AS at slow tempi, though these different phase behaviors are difficult to pull apart given the number of participants in each group.

At tempo changes, where interval correction is necessary, there is no difference between the two groups. Both show a similar overshoot pattern for increasing and decreasing tempo. This is surprising since the slope of the phase-correction response (the amount of correction at tap 1 after a perturbation, see Fig. 3) has been thought to reflect the strength of sensorimotor coupling.[24] Persons who stutter are theorized to have poor sensorimotor coupling, but in the present study, they did not differ from controls on this measure, even though the overshoot was tightly correlated with overall angular consistency within both groups. In the following sections, we consider the negative mean asynchrony, phase consistency, neural mechanisms, relationship with music, and fluency enhancement in more depth.

## *Negative mean asynchrony (i.e., mean phase angle)*

It appears that individuals who stutter as a group have a more negative phase angle than control participants, in both adolescents[17] and adults (trend from van De Vorst and Gracco;[27] the current study). However, when we examined phase angle as a function of stuttering severity, we found that it was the individuals with milder stuttering who had the most negative angles, while the more severe cases were closer to controls in their phase angle. Falk and colleagues[17] did not find this relationship, and proposed two subgroups within individuals who stutter based on their data: those who have extreme negative angles and those who have low consistency. If these subgroups exist, we might expect a negative relationship between phase angle and phase consistency within AS, but our data do not suggest this (Table 3).

In the general population, mean asynchronies are often related to musical training[26] and preferred or spontaneous rate.[25] For the current study, groups were matched on musical training, so this should not systematically affect our results. We are left with intrinsic rate: a more negative phase angle in the AS group should suggest a faster intrinsic rate.[25] The current study did not examine spontaneous rates, but Subramanian and Yairi did, asking individuals who stutter to tap at a "comfortable rate" and "as fast as possible." They found slower "comfortable" rates along with faster and more variable "fast" rates.[14] Such a pattern is complex but could indicate a shifted distribution for spontaneous rate. If individuals who stutter have a faster spontaneous rate, this could explain why they anticipated the beat and also why they performed less consistently at the slowest tempi (which are far from their preferred rate).

However, it may be that differences in spontaneous rate are not sufficient to explain this negative mean asynchrony in individuals who stutter. Instead, it could be related to a difference in inhibitory control, which some evidence suggests is affected for both children and adults who stutter.[38] For example, studies by Eggers *et al.* found that children who stuttered were much more likely to have a premature response on a go/no-go task and that their parents rated them lower on inhibition than did parents of children who did not stutter.[39,40] Similarly, Markett and colleagues found that stuttering adults took longer to respond to stop signals in a stop-signal reaction-time task,[41] and Subramanian and Yairi also reported shorter reaction times on a Stroop test.[14] Salmelin and colleagues even found early motor cortex activation during word production, indicating that there is also premature motor activity during speech in individuals who stutter.[42]

### *Measures of consistency and phase drift*

Phase consistency, or MRL (Fig. 2B), is the only measure that showed a significant interaction between tempo and group. One factor that could affect MRL in such a way is phase drift—if participants gradually change their relationship to the beat rather than oscillating around a single preferred angle, this would reduce MRL but not necessarily IRI. Indeed, Max and Yudman observed a greater amount of drift for individuals who stuttered than for controls in their synchronization-continuation task.[20] In our study, drift was more constrained than in a synchronization-continuation task by the constant presence of the metronome. The differences between groups in IRI and phase drift (Fig. 2) were both slightly more pronounced at slow tempi, though neither led to a statistically significant interaction. Using the circular measure of phase consistency, which combines both types of information, a significant interaction emerged. Increased variability in the nonspeech movements of individuals who stutter has been hotly contested,[16,19–22,43] and here we do find it in slower tempi. This variability suggests a less secure connection between auditory and motor systems in this population, contrary to what the tempo change data imply. This could be because we used larger tempo changes than the original study by Repp (we used changes of 75—150 ms, while Repp[24] used less noticeable changes of 5—25 ms). Thus, our tempo change measure might be less sensitive, leading to comparable results for both groups. Phase consistency, on the other hand, seems to be the most sensitive measure, and it is here that we detect differences in timing behavior between individuals who stutter and fluent speakers.

### *Possible neural mechanisms*

Timing perception and production, over intervals like those used in this study, is influenced by dopaminergic activity in the basal ganglia,[44] and it has been proposed that individuals who stutter have an excess or abnormal regulation of dopamine. Evidence for abnormal dopamine regulation includes the observation that stuttering can be attenuated with dopamine blockers,[45] and recent neuroimaging has shown atypical activation, connectivity, and morphology of the basal ganglia, whose functioning is intimately tied with dopamine.[46,47] However, timing does not depend solely on the basal ganglia. For example, it has been proposed that the lateral motor system is associated with the cerebellum and motor cortex and controls externally timed actions.[48–51] In contrast, the medial motor system is associated with basal ganglia, frontal, and parietal areas, and corresponds to internally generated timing.[48–51] This division of timing systems lines up well with what we know about neural activity in stuttering: there is less activity in basal ganglia[46,47,52] and the overactive cerebellum.[53,54] One possibility is that AS rely on the lateral timing system more than the medial timing system, and only in certain specific conditions does this difference in neural strategy translate to a difference in behavioral output.

### *Music and stuttering*

Finally, we observed a trend for AS to have lower scores on the music perception task, which is further evidence of perceptual differences between individuals who stutter and their fluent peers.[9,10] This was true despite the fact that both groups had similar amounts of music training, and that scores on this task were relatively high. In fact, if anything, individuals who stutter had an advantage in that those who had music training began earlier in life, which should be associated with better music perception and performance.[55,56] In addition, fluent individuals showed correlations between music and the IRI percent error, whereas music variables were not able to predict any aspects of synchronization in individuals who stuttered.

Alongside this difference in music perception was the intriguing finding that individuals who had more musical training also had lower stuttering severity (self-evaluated). The cause of this correlation is uncertain, and since the same correlation does not reach significance with the SLP's severity ratings, it must be taken cautiously and replicated. If the effect turns out to be robust, it might be because music training reduces stuttering severity, but it could also be that individuals who stutter have an underlying problem with time perception or other factors which predispose them to participate less in musical activities.[57]

### *Synchronizing speech and nonspeech*

Since speaking with a metronome stimulus is a fluency-enhancing condition for individuals who stutter and recent studies show that this may be useful in therapy,[58,59] it may seem paradoxical that individuals who stutter have less intact metronome synchronization compared with the general population. However, metronome synchronization and paced speech are two very different tasks, especially in terms of feedback: here, the participants did not have any self-produced auditory feedback to align with the metronome tick, whereas in a metronome-paced speaking situation, they have feedback from their own voice plus the metronome. Many fluency-enhancing techniques, like choral speech, altered feedback, and white noise, rely on increasing or manipulating any kind of auditory feedback rather than the rhythmicity of the stimulus.

## Conclusions

Using circular statistics and a tempo change paradigm, this study has provided evidence of atypical manual synchronization in individuals who stutter, along with music perception differences. The traditional measurement of the IRI error between the two groups produced at best a modest effect, which may explain why previous literature on manual synchronization in individuals who stutter has been inconsistent. In contrast, differences in phase angle were striking. Though the patterns of phase drift deserve further study, our results are consistent with the idea that stuttering is related to a generalized timing problem that affects nonspeech and speech. This may stem from deficits in a medial timing system in the brain responsible for internal timing. It is worth noting that this experiment effectively removed auditory feedback. When auditory feedback is added into the mix, the pattern of results may change.

The task in the current study, though harder than continuous synchronization to a single tempo, was still relatively easy. Perhaps clearer differences would be observed in tasks with subtler metronome changes, or by increasing the difficulty using bimanual tapping or syncopation. It could also be interesting to relate spontaneous tapping rates with synchronization performance in individuals who stutter, or even to probe other sensorimotor interactions such as visuomotor synchronization with these same methods. Finally, it is important to recognize that many neural processes could lead to differences in synchronization behavior, so it will be important in the future to look at brain—behavior relations and evaluate the timing network for speech and nonspeech in individuals who stutter.

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## Author contributions

A.S. designed and coded the experiment, tested participants, analyzed data, and wrote the first draft of the manuscript. M.D. analyzed data and contributed to the writing, especially the results section. D.S. assisted with the implementation of the experimental procedures and edited the manuscript. V.G. funded the work, gave input on the design and analysis, and edited the manuscript. All authors accept responsibility for the integrity of the data analyzed.

## Supporting information

Additional supporting information may be found in the online version of this article.

**Figure S1.** Phase consistency (MRL) and angle stabilization.

**Figure S2.** Distributions and mean resultant vectors by response after a tempo change.

**Figure S3.** Mean resultant vectors for all participants (18 participants per group).

**Figure S4.** Plot of the amount of phase drift (absolute slope of the steady-tempo portion).

**Table S1.** Correlation table for 18 participants per group

## Competing interests

The authors declare no competing interests.

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