Article 3: Faulty mechanisms of timekeeping in the beat-based form of congenital amusia

Pauline Tranchant, Isabelle Peretz

Faulty mechanisms of timekeeping in the beat-based form of congenital amusia

Pauline Tranchant[a,b] **& Isabelle Peretz**[a,b]

1 Département de psychologie, Université de Montréal, Montréal, Québec, Canada

2 International Laboratory for Brain, Music and Sound Research (BRAMS), Université de Montréal, Montréal, Québec, Canada

Keywords: beat, music, isochrony, timekeeping, movement, sensorimotor deficits

Abstract

Humans master the capacity to move in time with the beat of music. Yet some individuals show marked difficulties to synchronize simple body movements with musical beats. The causes of this phenomenon are still largely unexplained. Here we investigated internal timekeeping capacities, which are driving all rhythmic motor behaviors, in a group of eight beat-impaired and a group of 14 matched control participants. Beat-impaired cases were recruited for their poor ability to tap with the beat of music, and did not present any neurological, auditory or musical pitch-related deficits. Groups were compared for two finger tapping tasks: spontaneous production of regular sequences (no stimulus) and synchronization-continuation to a metronome spanning a large range of interval periods (225-1709 ms). Higher inter-tap variability was observed across tasks in the beat-impaired group. Synchronization was in addition characterized by unsuccessful matching of the fastest metronome's period and larger asynchronies between taps and tones of slow metronomes. A lower capacity to maintain the period of slow metronomes was as well observed during the continuation (i.e. after tones had stopped). Altogether these results indicate a low capacity for isochrony and limited rate flexibility in the beat-impaired group, and suggest a disruption of basic timekeeping mechanisms.

Introduction

Humans are particularly good at producing stable periodicities in synchrony with others (Kirschner & Tomasello, 2009; Richardson, Marsh, Isenhower, Goodman, & Schmidt, 2007; Su, 2014). This skill is biologically useful for cooperation (Cirelli, Einarson, & Trainor, 2014) and events prediction (Konvalinka, Vuust, Roepstorff, & Frith, 2010; van der Steen & Keller, 2013). It appears universally mastered to the exception of a few deviant individuals (Phillips-Silver et al., 2011). In the advent of a neurodevelopmental anomaly, synchronization skills can be disrupted. The disorder typically affect beat finding in music (Phillips-Silver et al., 2011; Sowiński & Dalla Bella, 2013) but it may also occur in synchronization to a metronome in which timing is strictly isochronous (Palmer, Lidji, & Peretz, 2014; Tranchant, Vuvan, & Peretz, 2016). Here we tested the basic mechanisms underlying the aptitude for isochrony in eight individuals who have been diagnosed with deficient musical beat finding, which is a form of congenital amusia.

The most common form of congenital amusia concerns the processing of the pitch structure of music, not isochrony (Hyde & Peretz, 2004; Phillips-Silver, Toiviainen, Gosselin, & Peretz, 2013). It is a neurodevelopmental disorder that affects the processing of musical pitch in both perception and production. It is associated to abnormal connectivity between the auditory cortex and inferior frontal cortex mostly in the right cerebral hemisphere (for a review see Peretz, 2016). The pitch disorder is hereditary (Peretz, Cummings, & Dubé, 2007) and molecular analyses are in progress in order to identify the responsible genes. Thus, pitch deafness is instructive regarding causal links between musical pitch, brain networks and genes. Likewise, impairments in isochrony represent a complementary and distinct chance to study the neurobiological foundations of musical rhythm.

The core mechanisms underlying musical beat arises from the aptitude to impose an isochronous grid to a rhythmic sequence. The aptitude for isochrony, where all intervals between events are equal like those of a steady metronome, rests on the interaction between acoustical cues and higher-level cognitive organization. One key feature of this high-level organization is anticipation. Humans tap in advance of metronome clicks by a few tens of milliseconds (Repp, 2005). This typical behavior is thought to compensate for the time lag introduced by sensory processes (Aschersleben, 2002; Aschersleben & Prinz, 1995; Aschersleben, Gehrke, & Prinz, 2001). Such anticipatory tendency is considered to be unique to humans and a few animal species, specifically those with the capacity for vocal learning (Patel, 2014; Patel, Iversen, Bregman, & Schulz, 2009), although recent findings with a sea lion (Cook, Rouse, & Wilson, 2013), a chimpanze (Hattori, Tomonaga, & Matsuzawa, 2013), and a bonobo (Large & Gray, 2015), who are not vocal learners, are challenging this view (Rouse, Cook, Large, & Reichmuth, 2016). In the two beat-impaired cases we have tested so far, normal anticipation of steady metronome's clicks was observed (Palmer et al., 2014).

Another key feature of the aptitude for isochrony is rate flexibility (McAuley, Jones, Holub, Johnston, & Miller, 2006). It is still unknown whether beat-impaired individuals lack flexibility, since they_were tested with a narrow range of isochronous rates, which lie around the optimal tempo of 500-600 ms between events (Baruch, Panissal-Vieu, & Drake, 2004; Moelants, 2002). The largest range tested with beat-impaired cases is 450-750 ms for which large individual differences are obtained (Sowiński & Dalla Bella, 2013). This range is still limited in regard to what adults can typically achieve when synchronizing taps with a metronome (Repp, 2005).

Here, we compare the performance of eight new beat-impaired cases with those of a control group in terms of anticipatory tapping (synchronization) and continuation to metronome-like stimuli in reference to their spontaneous tapping rate. First, based on previous studies (Palmer et al., 2014; Phillips-Silver et al., 2011; Sowiński & Dalla Bella, 2013; Tranchant et al., 2016) we predict that no difference will be observed for spontaneous tapping between the beat-impaired group and a group of matched control participants. This is assessed in Study 1. Second, we predict that synchronization and continuation performance may be normal initially around beat-impaired participants' spontaneous (comfortable) tapping rate but should deteriorate for stimulus rates rolling away from it. In study 2, we test to what extend synchronization-continuation degrades for rates that are distant from the individually defined spontaneous rate.

Methods

Participants

Eight beat-impaired cases (6 females, 2 males; mean age: 27.1 years, *SD* = 2.2) were matched for years of education and years of musical and dance training to 14 control participants (9 females, 5 males; mean age: 26.6 years, range 23-33 years, *SD* = 2.3). All participants were university students or recent graduates, and none had history of neurological or motor disorders. Six beat-impaired cases were selected on the basis of poor synchronization to music in our laboratory, two cases self-declared their inability to follow the beat in music. In order to confirm the presence or absence of a beat finding disorder, all participants' synchronization abilities were assessed with 20 songs, with the instruction to tap to the beat. Ten musical stimuli, which varied in genre and tempo, were presented twice over two blocks. Their duration was between 24 and 32 seconds, and the first and last five seconds of tapping were removed from the analyses. A detailed description of musical stimuli is provided in Table 1. We used circular statistics (Batschelet, 1981) to assess tapping synchronization to the beat.

We used the Rayleigh test to assess whether taps were period-matched with the beat period of each musical stimulus, after transforming taps into vectors on the circle. A significant Rayleigh test ($p < .05$) indicates success in period matching (i.e. inter-tap intervals are consistent with the stimulus inter-beat-interval). As can be seen in Figure 1, there was not overlap between the number of trials that were period-matched with the beat of the musical excerpts by beat-impaired cases and controls. To confirm the synchronization deficit, the circular variance was used to compare the performance of beat-impaired cases to normative scores obtained in our laboratory from 41 typical synchronizers (23 females, mean age: 26.6 years, $SD = 4.4$). The circular variance is a measure of consistency between inter-tap and inter-beat intervals, and is bounded by zero and one. A value close to zero indicates high consistency while a score close to one usually corresponds to a non-significant Rayleigh test and indicates a random distribution of vectors (one vector = one tap) around the circle. The circular variance score was positively skewed and therefore transformed using a log function (log score = -1*log circular variance), a higher score now indicates higher consistency. For each participant, the log-transformed values were averaged across the 20 trials, providing an index score of individual performance. All of our eight beat-impaired cases were below *SD* under the mean of the typical group (cut-off = 1.54), which confirms their poor beat-finding abilities.

[Insert Figure 1 here]

All participants were screened for the presence of other musical deficits than synchronization to music. Beat-impaired cases were evaluated with the Montreal Battery of Evaluation of Amusia (MBEA; Peretz, Champod, & Hyde, 2003). They obtained a normal melodic composite and rhythm score (Table 1; Vuvan et al., 2017). The melodic composite score is an averaged score for three tests (scale, contour and interval) assessing pitch processing in a melodic context. The rhythm test evaluates a participant's ability to judge whether two short melodies are similar or not, differences being created by manipulating the duration of two adjacent intervals while maintaining the beat structure. Contrary to what has been found in a previous study (Phillips-Silver et al., 2011), only two out of the eight cases scored two standard deviations below the mean on the Meter test, which assesses a participant's ability to judge whether short melodies have an underlying pattern of strong and weak beats that corresponds to either a march (One two One two) or a waltz (One two three One two three). Controls' musical abilities were tested with the Online Test of Amusia for which they all obtained normal scores (Peretz & Vuvan, 2017). Finally, all participants performed in the normal range for verbal working memory and non-verbal reasoning (Progressive Matrix and Digit Span tests from the WAIS-III Wechsler adult intelligence scale; Wechsler, Coalson & Raiford, 1997).

[Insert Table 1 here]

The research was approved by the local ethics committee at Université de Montréal and participants provided written informed consent. They received financial compensation for their participation.

Material and procedure

The design of the study is schematized in Figure 2. First, each participant was instructed to tap in silence (no stimulus) at her most comfortable rate, in a constant and regular fashion "as if she were a metronome". A sounded beep indicated when to start and when to stop, that is after 31 taps were made. Twenty such spontaneous tapping sequences were recorded over two sessions, with three in a row at the beginning of each session to measure the spontaneous period (P0).

[Insert Figure 2 here]

Next, each participant was invited to tap with a metronome (synchronization) and to continue thereafter (continuation). Before each trial, she was primed with a short version of the stimulus (10 tones) and instructed to listen without moving. Then, she was invited to tap in synchrony with 31 isochronous tones (440 Hz, 200 ms duration) with an inter-onset-interval (IOI) corresponding to her spontaneous tempo (P0) as computed from her own pace just before. She also tapped to tones series corresponding to P0 plus 50 ms (P0+50) and P0 minus 50 ms (P0-50). The order of P0+50 and P0-50 was counter-balanced between participants and inverted in the second testing session. After the tones had stopped, participants continued tapping at the same rate. A beep indicated to stop tapping after 31 continuation taps. Participants were instructed to maintain the tempo and regularity during the continuation.

The session ended by six synchronization-continuation trials of varying IOI and by seven spontaneous tapping sequences. Stimulus IOIs were fixed here and consisted of 225, 337, 506, 759, 1139, and 1709 ms as in (McAuley et al., 2006). A short version (10 tones) of the stimulus was presented before each synchronization-continuation trial for preparation. The stimuli were presented in a descending (fast to slow) or ascending (slow to fast) order, which was counter-balanced between participants and inverted in the second session. The spontaneous sequences were performed before, between and after synchronization-continuation sequences (see Figure 2).

Each participant was tested in all conditions twice over two separate sessions, with a minimum of six days between sessions. Each session lasted about 30 minutes. Taps were made on a square force sensitive resistor (3.81 cm, Interlink FSR 406) placed on a table in front of the participant. The resistor was connected to an Arduino Duemilanove transmitting timing information to a PC (HP ProDesk 600 G1, Windows 7) via the serial USB port. Tap times were recorded and stimuli were generated with a customized program in MAX/MSP (Cycling' 74) at a sound level of 88 dB SPL through headphones (DT 770 PRO, Beyerdynamics).

Data Analysis

For each participant, there were 20 spontaneous tapping sequences, 18 synchronization-continuation sequences, and four (two slow, two fast) accessible tapping rates. The data were analyzed in Matlab (R2014b, Mathworks) with linear and circular methods, using the CircStat Toolbox (Berens, 2009). Statistical analyses were performed with linear mixed-effects models, using the "lme4" package (Bates, Maechler, Bolker, & Walker, 2015) in R (R Core Team, 2015). Plotted residuals indicate unequal variance across the range of predicted values for the circular variance. Therefore, circular variance scores were transformed using the reverse log function (new score = -1*log(old score), the higher the score the better the consistency). The models include a random term for Participant to account for repeated measures. Factor IOI (Study 2) was centered and scaled using the *scale* function in R before running mixed-effects statistical models, so that factors Group and IOI were on comparable scales. We used mixed-effects models rather than ANOVA because they do not require prior averaging of the data and have more statistical power in case of unbalanced designs such as different group sizes. Corresponding p-values and degrees of freedom are computed from the Satterthwaite approximations, using the "lmerTest" package (Kuznetsova, Brockhoff, & Christensen, 2017) in R. For post-hoc analyses, pairwise comparisons were performed with the "lsmeans" package (Lenth, 2016) in R, which computes least-square rather than arithmetic means to account for unbalanced designs.

Study 1: Internal timekeeping

The goal of Study 1 was to test participants' regularity in their spontaneous rate of tapping. To this aim, the first three tapping sequences collected at the beginning of each session were analyzed in order to avoid potential carry-over effects of synchronization-continuation rates on spontaneous rate.

Results

There were large variations in mean spontaneous inter-tap-interval (ITI), ranging from 471ms to 996ms in the beat-impaired group (M = 754ms) and from 391 ms to 1214 ms in the control group (M = 670ms; Figure 3A); the group difference was not significant, β = -84.15, SE = 66.23, $t(20)$ = -1.42, p = .22. In contrast, the two groups differed in regularity (Figure 3B). The coefficient of variation (CV), which corresponds to the standard deviation of the ITIs divided by the mean ITI of a sequence, indicated higher variability, hence poorer regularity in the beat-impaired (M = 0.074) as compared to the control group (M = 0.052). The difference in CV between the two groups reached significance, β = - .022, SE = .0046, $t(20)$ = -4.63, p < .001.

This difference between groups contrasts with previous studies. For example, in Sowiński & Dalla Bella (2013) the variability of spontaneous tapping in four beat-impaired participants (synchronization profile comparable to our beat-impaired group) was described as normal. This discrepancy may be due to the larger amount of data considered in the present study. To check for that, we compared our groups on the spontaneous sequence showing the lowest CV from the two first produced sequences (instead of six sequences), following the procedure of Sowiński & Dalla Bella. This analysis revealed no statistical difference between groups, $t(14) = -1.75$, $p = 0.10$ (Welch's two-sample t-test). Thus, higher statistical power (the six sequences were individually considered in the statistical model above) likely contributed to the finding of larger inter-tap variability in our beat-impaired group.

Regularity was unrelated to the individual spontaneous rate. There was no significant correlation between CV and spontaneous rate; $r(20) = .26$, $p = .23$ across all participants and $r(6) = .28$, $p = .50$ for the beat-impaired group only. The CV remained higher in the beat-impaired group ($M = 0.088$) as compared to the control group ($M = 0.057$) throughout the sessions, considering the averaged CV over the other 14 sequences performed in silence, $\beta = -.030$, $SE = .006$, $t(20) = -5.03$, $p < .0001$.

[Insert Figure 3 here]

Note that in both groups, the average spontaneous rate (745 and 670ms) was slower than the standard 500-600 ms mentioned in the introduction (e.g. Moelants, 2002). Thus, individually produced spontaneous rate (P0) needs to be taken into consideration when evaluating synchronization abilities, which was the goal of Study 2.

Study 2: Flexibility

The higher variability in spontaneous tapping observed in beat-impaired individuals as compared to controls suggests that basic timekeeping mechanisms are impaired in the beat-related form of congenital amusia. Providing an external aid from metronome sounds may assist timekeeping and hence decrease variability in tapping. To be effective as an aid, it is likely that the metronome rate should be proximal to the spontaneous rate (P0). Furthermore, continuation (i.e. without the external aid) is expected to be disrupted, especially for stimulus rates distant from P0. The goal of Study 2 was to examine these predictions.

Results

The individual P0 was computed from the sequence with highest regularity (lowest CV) over the three first spontaneous sequences of each session. P0 ranged from 553 to 882 ms in the beat-impaired group and from 426 to 983 ms in the control group, with no difference between groups, $\beta = -84$, $SE = 66$, $t(20) = -1.27$, $p = .22$.

The CV (synchronization) of Inter-Tap-Intervals (ITI) for the Inter-Onset-Intervals (IOI) set to individual P0 and P0±50ms was again higher in the beat-impaired ($M = .068$) than in the control group ($M = .052$), $\beta = -.016$, $SE = .0031$, $t(20) = -5.15$, $p < .0001$. However, the CV does not capture anticipation and accuracy of synchronization because it does not take the stimulus into account. For example, a sequence of taps could be highly regular (low CV) but not synchronized with the tones. Thus, the log transformed circular variance (see Data Analysis) was considered here as the main variable. These scores indicate lower consistency in the beat-impaired compared to the control group, $\beta = 0.70$, $SE = .19$, $t(20) = 3.77$, $p = .0012$

(Figure 4). Furthermore, the beat-impaired group anticipated the tones in an anomalously large degree of magnitude. The asynchronies between taps and tones, computed by subtracting the closest tone onset time from the tap time, indicated a negative trend with a tap occurring before the tone onset, which was much larger in the beat-impaired compared to the control group, $\beta = -48$, $SE = 11$, $t(20) = -4.15$, $p < .001$ (Figure 4). Thus, taps tended to be further away from the tones in the beat-impaired compared to the control group when tapping to tones at the individual spontaneous tempo.

[Insert Figure 4 here]

Continuation performance was similar to synchronization, with higher CV (lower regularity) in the beat-impaired ($M = .069$) compared to the control group ($M = .056$), $\beta = -.013$, $SE = .0045$, $t(20) = -3.01$, $p < .01$. Yet, the ability to maintain the stimulus rate, as measured by the distance between the mean ITI and the stimulus IOI (i.e. continuation error), did not differ between groups, $\beta = -12$, $SE = 9$, $t(20) = -1.39$, $p = .18$. Thus, higher CV in the beat-impaired group was due to poor regularity rather than a constant drift towards a faster or slower tempo than the stimulus.

To summarize, the results show that even at a participant's most comfortable rate and with the external aid of isochronous tones, tapping performance remains poorer in the beat-impaired group as compared to controls. In what follows, we assessed to what extent less comfortable stimulus rates disrupted performance.

The synchronization-continuation performance for the six pacing rates in the range of 225-1709 ms is presented in Figure 5. The results indicate limited flexibility in the beat-impaired group, particularly at fast paces. Actually, for the fastest pace (IOI = 225 ms), the Rayleigh test revealed a failure to period-match ($p > .05$) in six of the eight beat-impaired cases whereas none of the participants in the control group failed the Rayleigh test at that rate. Note that the failure to tap regularly does not seem to arise from biomechanical or motor limitations because beat-impaired participants were producing fast rates when tapping to fast stimuli (see Table 2).

[Insert Figure 5 here]

[Insert Table 2 here]

Because sequences were unsuccessfully period-matched with the fastest stimulus rate (IOI = 225 ms) in most beat-impaired individuals, this rate was not included in subsequent analyses of synchronization consistency and accuracy. Synchronization difficulties were also observed for the second fastest rate (337 ms; Figure 5); a failure to period-match with that stimulus rate was observed in three beat-impaired cases. These failed sequences were not included in the following analysis of synchronization consistency and accuracy.

As can be seen in Figure 5, synchronization consistency, measured by log-transformed circular variance, was generally lower in the beat-impaired group as compared to the control group, $\beta = .90$, $SE = .14$, $t(20) = 6.52$, $p = <.0001$, with an effect of Stimulus Rate, $\beta = .19$, $SE = .040$, $t(192) = 4.67$, $p = <.0001$, and no interaction between Group and Stimulus Rate factors, $\beta = -.089$, $SE = .080$, $t(192) = -1.11$, $p = .27$. Post-hoc comparisons for the effect of Stimulus Rate (with Bonferroni-holm p-value adjustment) showed lower consistency in both groups for 337ms compared to 759, 1139 and 1709 ms (all $p < .05$) and for 506 ms compared to 1139ms ($p < .01$) across groups.

Anticipation of tone onsets was observed in both groups: mean asynchronies were negative for 84% and 85% of the sequences in the beat-impaired and control groups, respectively. Thus, taps again tended to precede tones onsets in both groups, but again to an anomalously larger degree in the beat-impaired group for slow metronome rates. A summary of mean asynchronies by rate for each group is provided in Table 3. There was an effect of Group on asynchronies (synchronization accuracy), $\beta = -40$, $SE = 14$, $t(20) = -2.89$, $p = .00091$, an effect of Stimulus Rate, $\beta = 27$, $SE = 2.66$, $t(192) = 10.13$, $p < .0001$, and an interaction between Group and Stimulus Rate factors, $\beta = -29$, $SE = 5.31$, $t(192) = -5.54$, $p < .0001$. The distance between taps and tones increased with the IOI in the beat-impaired group but not in the control group (Figure 5). There was no group difference for 337 and 506 ms (both $p > .05$ by post-hoc comparisons with Bonferroni-holm p-value adjustment), but significantly larger asynchronies in the beat-impaired group for 759 ($p = .028$), 1039 ($p = .0038$), and 1709 ms ($p < .0001$). Because the mean asynchrony was negative (see Table 3), larger asynchronies for slow rates in the beat-impaired group indicate a tendency to underestimate the stimulus IOI.

[Insert Table 3 here]

The CV of ITIs during continuation was higher in the beat-impaired compared to the control group, $\beta = -.021$, $SE = .0031$, $t(20) = -6.58$, $p = <.0001$, with no effect of Stimulus Rate or of Tempo (both $p > .22$). Thus, continuation was generally less regular in the beat-impaired than in the control group, like observed in spontaneous and synchronization tapping. Continuation error was computed by subtracting the mean ITI from the stimulus IOI (in magnitude). While the error tended to increase with larger IOIs (slower rates) in both groups, the effect was generally larger in the beat-impaired group (Figure 5), as supported by an interaction between Group and Stimulus Rate factores, $\beta = -42$, $SE = 10$, $t(216) = -4.36$, $p < 0.0001$. The two groups did not differ for the fast rates (225, 337, 506 and 759ms rates; all $p > .05$) but did so for the slow rates (1139 ms; $p = .0028$ and 1709 ms; $p < .0001$). Thus, contrary to predictions, beat-impaired participants were as capable as controls to maintain fast and moderate rates after the stimulus had stopped. Of note is that, because the number of taps is fixed, the length of continuation sequences increases with slower rates. We checked whether keeping only the first half of continuation taps for the 1139 and 1709 ms stimuli suppressed the difference between groups, and it did not.

Note that all the analyses presented above were performed with groups of similar sizes, by considering eight control participants instead of 14. All results are maintained except for the larger asynchronies during synchronization at 759 ms IOI in the beat-impaired group.

Discussion

The main finding of the present study is that individuals with anomalous difficulties to tap to the beat of music have more basic problems with timekeeping mechanisms. We found a lower capacity for isochrony (higher inter-tap variability) in the beat-impaired group across conditions. We also found evidence that rate flexibility is limited in the beat-impaired group: larger negative asynchronies as well as poor tempo retention were observed for slower metronome rates, and a striking limitation was observed for synchronization to fast rates.

Our results can be interpreted within a nonlinear dynamical approach to beat-based coordination of motor actions (Drake, Jones, & Baruch, 2000; Large & Jones, 1999; Loehr, Large, & Palmer, 2011; McAuley, 2010; McAuley et al., 2006). In this approach, synchronization to a regular beat is considered to rely on an internal oscillator, which is capable to generate an intrinsic beat and to adapt the period of this beat to match that of the stimulus. The intrinsic period of the oscillator is captured by spontaneous tapping, while period adaptation is measured by synchronization-continuation, with higher adaptation demands for extreme fast and slow rates. The operation of this internal oscillator may be the faulty mechanism in the beat-impaired cases.

First, higher inter-tap variability (CV) across conditions reveals a disrupted capacity of the oscillator for isochrony. In particular, high variability during spontaneous tapping indicates that the impairment goes beyond poor sensorimotor coupling with external signals. This finding contrasts with prior reports of beat impairments (Phillips-Silver et al., 2011; Sowiński & Dalla Bella, 2013; Tranchant et al., 2016). In Phillips-Silver et al. (2011), the variability of spontaneous whole-body bouncing movement in one beat-impaired case (Mathieu) was described as normal, although comparison to the control group was not considered. In Tranchant et al. (2016), the variability of spontaneous bouncing as well as hands clapping did not differ between beat-impaired and control groups. It is possible that differences between movement forms contributed to the discrepancy between prior and present studies. The beat-impaired case (Mathieu) from Phillips-Silver et al. (2011) was later tested by Palmer et al. (2014) with finger tapping. His inter-tap variability (CV) in spontaneous production was then above one standard deviation from the mean of the control group, and very close to findings in our beat-impaired group (0.072 in Mathieu compared to a mean of 0.074 in our beat-impaired group). Finally, higher statistical power in the present study, as for example compared to Sowiński & Dalla Bella (2013), may have contributed to highlight the difference between groups in spontaneous tapping.

Second, limited rate flexibility in the beat-impaired group reflects poor period adaptation of the oscillator, for rates that are beyond the optimal range. In other terms, the range for which period adaptation is optimal is narrower in beat-impaired cases. We indeed observed poor synchronization and/or continuation performances at extreme slow and fast rates. A severe difficulty to synchronize with the fastest stimulus rate (225 ms) could not be attributed to pure motor or biomechanical limitations, because produced periods were close or sometimes even shorter than the target. The limitation thus likely emerges from faulty period adjustment mechanisms. For slow rates, beat-impaired cases showed a lower capacity to use error signals for synchronization, as indicated by larger asynchronies, and showed low period stability in the absence of the external aid, as indicated by large deviations from the target period in continuation. Altogether, these findings point toward faulty period adaptation mechanisms at extreme slow and fast rates.

The absence of such a difference between groups for fast rates, as found here, may seem against this conclusion. For example, McAuley et al. (2006) observed larger continuation errors with extreme slow *and* fast rates, as compared to older children and to adults. These authors concluded of a narrower range of optimal adaptation in young children. Yet continuation errors in children below eight were much larger for slow as compared to fast rates in that study (see Figure 7) and were very similar to findings in our beat-impaired group. It is thus possible that the limited size of our groups was responsible for the difference between groups not being significant for the fastest rates.

Additional support for the faulty period adaptation hypothesis also comes from previous studies of beat impairments. For example, Mathieu (Phillips-Silver et al., 2011) showed poor adjustment of bouncing movements for a gradual 10% tempo change applied to a musical excerpt, but not for a 20% tempo change. However, in that study period matching was not precisely assessed (only the direction of speed change) and comparison to a control group was not considered. Thus, it is possible that adaptation to gradual tempo changes was even more severe than the findings suggest. Palmer et al. (2014) further assessed Mathieu's tapping to sudden (unpredictable) perturbations in otherwise isochronous sequences of tones. Perturbations consisted in small increases or decreases (3%, 8% or 15%) from the baseline of 500 ms IOI (i,e. by 15, 40 or 75ms). The number of taps required to return, after the perturbation, to baseline alignment with tones was longer in Mathieu than in controls. In addition, this difference between Mathieu and the control group was successfully captured by a damped harmonic oscillator model. The findings of Palmer et al. (2014) thus fit the hypothesis of poor period adaptation of an internal oscillator in the beat-impaired condition.

A challenge for future research will be to identify the neural bases underlying beat impairments. In the nonlinear dynamical approach, temporal coordination is supported by self-sustained ongoing neural oscillations which entrain to the pulse of an external rhythmic signal (for a recent review see Haegens & Zion Golumbic, 2018). Which of the brain areas are generating and/or supporting this mechanism is not fully understood yet. In a meta-analysis of 43 functional neuroimaging studies, Chauvigné, Gitau & Brown (2014) contrasted regions showing activations during spontaneous versus synchronization tapping. They found a dissociation between two subcortical structures frequently associated to motor timing: the cerebellum (e.g. Buonomano & Mauk, 1994; Paquette, Fujii, Li, & Schlaug, 2017; Penhune, Zatorre, & Evans, 1998) and basal ganglia (e.g. Hausdorff, Cudkowicz, Firtion, Wei, & Goldberger, 1998; Schwartze, Keller, Patel, & Kotz, 2011). This analysis revealed that while basal ganglia seem to be important for the two types of motor tasks, the cerebellum seems involved in synchronization tapping only. Functional imaging studies of beat impairments are a rare chance to provide causal links between brain regions and behavior. In particular, because a poor capacity for isochrony was found for both spontaneous and synchronization tapping, we predict that anomalies related to the basal ganglia will be detected in a beat-impaired group.

To conclude, we showed that deficient synchronization to music can be traced back to human's core timekeeping mechanisms. The hypothesis of a faulty internal oscillator provides a useful model to interpret the findings. Deficiencies in timekeeping and temporal coordination are a rare chance to better understand human timing, which is essential for numerous human activities, including dancing, music making or even speech. For example, smooth turn-taking in conversations requires a form of temporal coordination thought to rely on synchronization between oscillators in the brains of people taking part in the conversation (Wilson & Wilson, 2005). Future studies of the beat-impaired brain should advance our understanding of the neural circuits that are essential for beat finding and timekeeping in general.

Acknowledgements

We would like to thank Mailis Rodrigues for her help with task programming.

Funding

This work was supported by the Natural Sciences and Engineering Research Council of Canada, and the Canada Research Chairs program.

Disclosure statement

The authors declare no competing interests

References

Aschersleben, G. (2002). Temporal Control of Movements in Sensorimotor Synchronization. *Brain and Cognition*, *48*, 66–79. http://doi.org/10.1006/brcg.2001.1304

Aschersleben, G., & Prinz, W. (1995). Synchronizing actions with events: The role of sensory information. *Perception & Psychophysics*, *57*(3), 305–317. http://doi.org/10.3758/BF03213056

Aschersleben, G., Gehrke, J., & Prinz, W. (2001). Tapping with peripheral nerve block : A role for tactile feedback in the timing of movements. *Experimental Brain Research*, *136*(3), 331–339.

Baruch, C., Panissal-Vieu, N., & Drake, C. (2004). Preferred perceptual tempo for sound sequences: Comparison of adults, children, and infants. *Perceptual and Motor Skills*, *98*(1), 325–339. http://doi.org/10.2466/pms.98.1.325-339

Bates, D., Maechler, M., Bolker, B. M., & Walker, S. C. (2015). Fitting linear mixed-effects models using lme4. *Journal of Statistical Software*, *67*(1), 1–48.

Batschelet, E. (1981). Circular statistics in biology. Academic Press, New York.

Berens, P. (2009). CircStat: a MATLAB toolbox for circular statistics. *Journal of Statistical Software*, *31*(10), 1-21.

Buonomano, D. V., & Mauk, M. D. (1994). Neural-network model of the cerebellum - temporal discrimination and the timing of motor-responses. *Neural Computation*, *6*(1), 38–55.

Chauvigne, L. A. S., Gitau, K. M., & Brown, S. (2014). The neural basis of audiomotor entrainment: an ALE meta-analysis. *Frontiers in Human Neuroscience*, *8*, 778. http://doi.org/10.3389/fnhum.2014.00776

Cirelli, L. K., Einarson, K. M., & Trainor, L. J. (2014). Interpersonal synchrony increases prosocial behavior in infants. *Developmental Science*, *17*(6), 1003–1011.

Cook, P., Rouse, A., & Wilson, M. (2013). A california sea lion (*zalophus californianus*) can keep the beat: Motor entrainment to rhythmic auditory stimuli in a non vocal mimic. *Journal of Comparative Psychology*, *127*(4), 412–427. http://doi.org/10.1037/a0032345

Drake, C., Jones, M. R., & Baruch, C. (2000). The development of rhythmic attending in auditory sequences: attunement, referent period, focal attending. *Cognition*, *77*(3), 251–288. http://doi.org/10.1016/S0010-0277(00)00106-2

Haegens, S., & Zion Golumbic, E. (2018). Rhythmic facilitation of sensory processing: A critical review. *Neuroscience and Biobehavioral Reviews*, *86*, 150–165. http://doi.org/10.1016/j.neubiorev.2017.12.002

Hattori, Y., Tomonaga, M., & Matsuzawa, T. (2013). Spontaneous synchronized tapping to an auditory rhythm in a chimpanzee. *Scientific Reports*, *3*, 1566. http://doi.org/10.1038/srep01566

Hausdorff, J. M., Cudkowicz, M. E., Firtion, R., Wei, J. Y., & Goldberger, A. L. (1998). Gait variability and basal ganglia disorders: Stride-to-stride variations of gait cycle timing in Parkinson's disease and Huntington's disease. *Movement Disorders : Official Journal of the Movement Disorder Society*, *13*(3), 428–437. http://doi.org/10.1002/mds.870130310

Hyde, K. L., & Peretz, I. (2004). Brains that are out of tune but in time. *Psychological Science*, *15*(5), 356–360.

Kirschner, S., & Tomasello, M. (2009). Joint drumming: Social context facilitates synchronization in preschool children. *Journal of Experimental Child Psychology*, *102*, 299–314.

Konvalinka, I., Vuust, P., Roepstorff, A., & Frith, C. D. (2010). Follow you, follow me: Continuous mutual prediction and adaptation in joint tapping. *Quarterly Journal of Experimental Psychology*, *63*(11), 2220–2230. http://doi.org/10.1080/17470218.2010.497843

Kuznetsova, A., Brockhoff, P. B., & Christensen, R. H. B. (2017). lmerTest Package: Tests in Linear Mixed Effects Models. *Journal of Statistical Software*, *82*(13), 1–26.

Large, E. W., & Gray, P. M. (2015). Spontaneous tempo and rhythmic entrainment in a bonobo (pan paniscus). *Journal of Comparative Psychology*, *129*(4), 317–328. http://doi.org/10.1037/com0000011

Large, E. W., & Jones, M. R. (1999). The dynamics of attending: How people track time-varying events. *Psychological Review*, *106*(1), 119–159. http://doi.org/10.1037/0033-295X.106.1.119

Lenth, R. V. (2016). Least-squares means: The R package lsmeans. *Journal of Statistical Software*, *69*(1), 1–33.

Loehr, J. D., Large, E. W., & Palmer, C. (2011). Temporal coordination and adaptation to rate change in music performance. *Journal of Experimental Psychology: Human Perception and Performance*, *37*(4), 1292–1309.

McAuley, J. D. (2010). Tempo and rhythm. In M. R. Jones, R. R. Fay, & A. N. Popper (Eds.), *Music perception. Springer handbook of auditory research* (Vol. 36, pp. 165–199). New York.

McAuley, J. D., Jones, M. R., Holub, S., Johnston, H. M., & Miller, N. S. (2006). The time of our lives: Life span development of timing and event tracking. *Journal of Experimental Psychology: General*, *135*(3), 348. http://doi.org/10.1037/0096-3445.135.3.348

Moelants, D. (2002). Preferred tempo reconsidered. In C. Stevens, D. Burnham, G. McPherson, E. Schubert, & J. Renwick (Eds.), *Proceedings of the 7th International Conference on Music Perception and Cognition* (pp. 580–583). Sydney, Australia: Causal Productions.

Palmer, C., Lidji, P., & Peretz, I. (2014). Losing the beat: Deficits in temporal coordination. *Philosophical Transactions of the Royal Society B: Biological Sciences*, *369*(1658), 20130405.

Paquette, S., Fujii, S., Li, H. C., & Schlaug, G. (2017). The cerebellum's contribution to beat interval discrimination. *Neuroimage*, *163*, 177–182. http://doi.org/10.1016/j.neuroimage.2017.09.017

Patel, A. D. (2014). The evolutionary biology of musical rhythm: Was darwin wrong? *Plos Biology*, *12*(3), e1001821. http://doi.org/10.1371/journal.pbio.1001821

Patel, A. D., Iversen, J. R., Bregman, M. R., & Schulz, I. (2009). Experimental evidence for synchronization to a musical beat in a nonhuman animal. *Current Biology*, *19*, 827–830. http://doi.org/10.1016/j.cub.2009.03.038

Penhune, V. B., Zatorre, R. J., & Evans, A. C. (1998). Cerebellar contributions to motor timing: A PET study of auditory and visual rhythm reproduction. *Journal of Cognitive Neuroscience*, *10*(6), 752–765.

Peretz, I. (2016). Neurobiology of congenital amusia. *Trends in Cognitive Sciences*, *20*(11), 857–867. http://doi.org/10.1016/j.tics.2016.09.002

Peretz, I., & Vuvan, D. T. (2017). Prevalence of congenital amusia. *European Journal of Human Genetics : EJHG*, *25*(5), 625–630. http://doi.org/10.1038/ejhg.2017.15

Peretz, I., Champod, A. S., & Hyde, K. (2003). Varieties of musical disorders. The Montreal

Battery of Evaluation of Amusia. *Annals of the New York Academy of Sciences*, *999*(1), 58–75.

Peretz, I., Cummings, S., & Dubé, M. P. (2007). The genetics of congenital amusia (tone deafness): a family-aggregation study. *The American Journal of Human Genetics*, *81*(3), 582–588.

Phillips-Silver, J., Toiviainen, P., Gosselin, N., & Peretz, I. (2013). Amusic does not mean unmusical: Beat perception and synchronization ability despite pitch deafness. *Cognitive Neuropsychology*, *30*(5), 311–331. http://doi.org/10.1080/02643294.2013.863183

Phillips-Silver, J., Toiviainen, P., Gosselin, N., Piché, O., Nozaradan, S., Palmer, C., & Peretz, I. (2011). Born to dance but beat deaf: A new form of congenital amusia. *Neuropsychologia*, *49*(5), 961–969. http://doi.org/10.1016/j.neuropsychologia.2011.02.002

Repp, B. H. (2005). Sensorimotor synchronization: A review of the tapping literature. *Psychonomic Bulletin & Review*, *12*(6), 969–992.

Richardson, M. J., Marsh, K. L., Isenhower, R. W., Goodman, J. R. L., & Schmidt, R. C. (2007). Rocking together: Dynamics of intentional and unintentional interpersonal coordination. *Human Movement Science*, *26*(6), 867–891. http://doi.org/10.1016/j.humov.2007.07.002

Rouse, A. A., Cook, P. F., Large, E. W., & Reichmuth, C. (2016). Beat keeping in a sea lion as coupled oscillation: Implications for comparative understanding of human rhythm. *Frontiers in Neuroscience*, *10*. http://doi.org/10.3389/fnins.2016.00257

Schwartze, M., Keller, P. E., Patel, A. D., & Kotz, S. A. (2011). The impact of basal ganglia lesions on sensorimotor synchronization, spontaneous motor tempo, and the detection of tempo changes. *Behavioural Brain Research*, *216*(2), 685–691. http://doi.org/10.1016/j.bbr.2010.09.015

Sowiński, J., & Dalla Bella, S. (2013). Poor synchronization to the beat may result from deficient auditory-motor mapping. *Neuropsychologia*, *51*(10), 1952–1963. http://doi.org/10.1016/j.neuropsychologia.2013.06.027

Su, Y. H. (2014). Audiovisual beat induction in complex auditory rhythms: Point-light figure movement as an effective visual beat. *Acta Psychologica*, *151*, 40–50.

Tranchant, P., Vuvan, D. T., & Peretz, I. (2016). Keeping the beat: A large sample study of bouncing and clapping to music. *Plos One*, *11*(7), e0160178.

van der Steen, M. C. M., & Keller, P. E. (2013). The ADaptation and Anticipation Model (ADAM) of sensorimotor synchronization. *Frontiers in Human Neuroscience*, *7*, 253. http://doi.org/10.3389/fnhum.2013.00253

Vuvan, D. T., Paquette, S., Mignault Goulet, G., Royal, I., Felezeu, M., & Peretz, I. (2017). The Montreal Protocol for Identification of Amusia. *Behavior Research Methods*, 1–11. http://doi.org/10.3758/s13428-017-0892-8

Wilson, M., & Wilson, T. P. (2005). An oscillator model of the timing of turn-taking. *Psychonomic Bulletin & Review*, *12*(6), 957–968.

Article 3: Tableaux et Figures

Tableau I. A3: Résultats à la MBEA des participants avec trouble de la synchronisation.

Table 1. MBEA scores. Individual scores of the 8 beat-impaired cases for the melodic (scale, contour, interval) tests, the rhythm and meter test of the Montreal Battery of Evaluation of Amusia. Scores below cut-off (2 *SD* below the mean; Vuvan et al., 2017) are in bold.

Participant	Melodic Composite Score	Rhythm	Meter
1	26.0	22	25
2	24.7	26	24
3	26.3	28	20
4	25.0	26	19
5	24.3	25	21
6	23.7	27	13
7	23.0	26	16
8	25.3	25	23

Tableau II. A3: Intervalle entre les tapes - tempo rapide.

Table 2. ITIs when tapping to the fastest stimulus IOI.

	ITI (ms)
Beat-impaired cases:	
1	206
2	233
3	254
4	301
5	199
6	233
7	231
8	210
Controls:	
M (range)	225 (223-227)

Tableau III. A3: Asynchronie moyenne

Table 3. Mean asynchrony between taps and tone onsets. A negative value indicates that taps precede tone onsets.

Group:	IOI:	337 ms	506 ms	759 ms	1139 ms	1709 ms
Beat-impaired	***M (SD)***	-16 (32)	-49 (39)	-78 (67)	-101 (67)	-136 (129)
Control	***M (SD)***	-19 (21)	-39 (30)	-33 (30)	-46 (31)	-43 (61)

Figure I. A3: Nombre d'essais avec alignement de période.

Figure 1. Number of period-matched trials (out of 20) per participant. Each participant is indicatd by a dot.

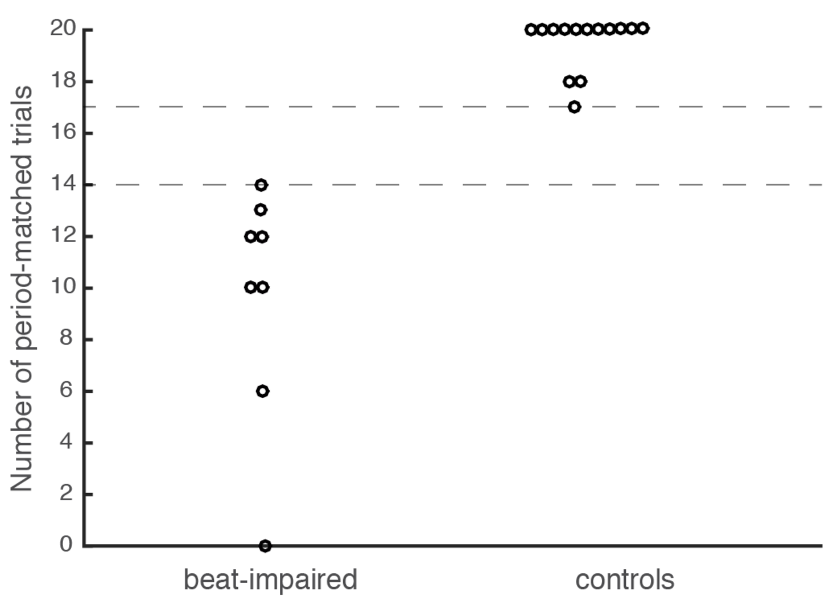

Figure II. A3: Description de la procédure

Figure 2. Procedure. S.T. = Spontaneous Tapping

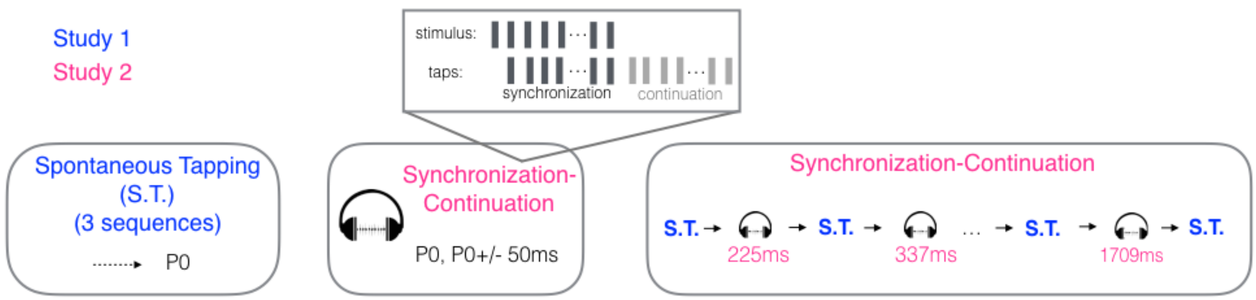

Figure III. A3: Tapping spontané

Figure 3. Spontaneous tapping. Each cross represents the mean score of one individual.

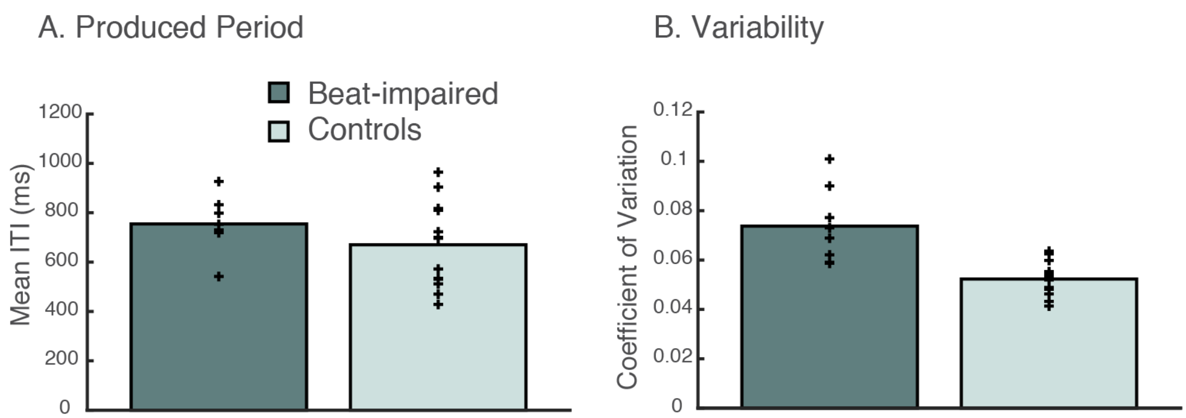

Figure IV. A3: Synchronisation-continuation au tempo spontané

Figure 4. Synchronization-continuation at participant's spontaneous tempo (P0). The average score obtained for six sequences (three per session) for each participant is represented by a dot. Bar graphs represent arithmetic means. For synchronization consistency, the higher the score the better the performance. For synchronization accuracy and continuation error, the lower the score the better the performance.

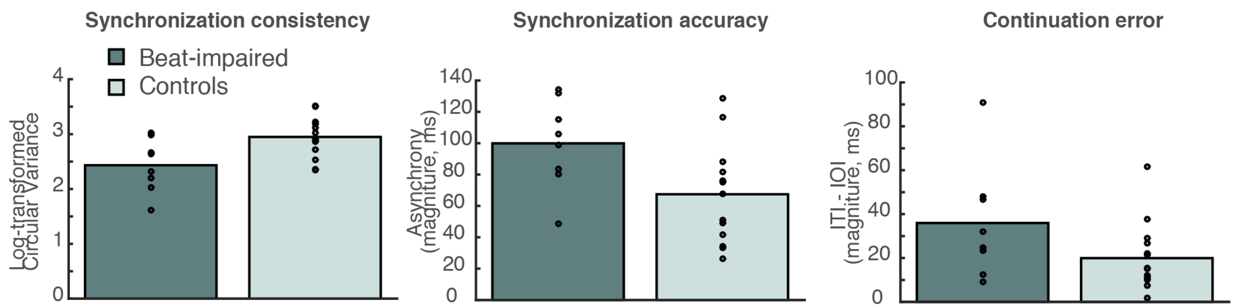

Figure V.	A3: Synchronisation-continuation aux tempi fixes.

Figure 5. Synchronization-continuation. IOIs of 225 (continuation only), 337, 506, 759, 1139, and 1709 ms. The graphs show arithmetic means and standard error bars (corrected for repeated measures).