



Research Report

The co-occurrence of pitch and rhythm disorders in congenital amusia



Marie-Élaine Lagrois^{a,b} and Isabelle Peretz^{a,b,*}

^a International Laboratory for Brain, Music, and Sound Research (BRAMS), Montréal, Québec, Canada

^b Department of Psychology, University of Montreal, Quebec, Canada

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ABSTRACT

The most studied form of congenital amusia is characterized by a difficulty with detecting pitch anomalies in melodies, also referred to as pitch deafness. Here, we tested for the presence of associated deficits in rhythm processing, beat in particular, in pitch deafness. In Experiment 1, participants performed beat perception and production tasks with musical excerpts of various genres. The results show a beat finding disorder in six of the ten assessed pitch-deaf participants. In order to remove a putative interference of pitch variations with beat extraction, the same participants were tested with percussive rhythms in Experiment 2 and showed a similar impairment. Furthermore, musical pitch and beat processing abilities were correlated. These new results highlight the tight connection between melody and rhythm in music processing that can nevertheless dissociate in some individuals.

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1. Introduction

Musical engagement is ubiquitous and emerges early in life. As soon as they are born, humans respond to abstract properties of musical pitch and time structure, such as changes in tonal key (Perani et al., 2010) and disruptions of musical beat (Winkler, Háden, Ladinig, Sziller, & Honing, 2009). Infants move spontaneously to music (Zentner & Eerola, 2010) and show enhanced pro-social behavior when moved in

synchrony with music (Cirelli, Einarson, & Trainor, 2014). In this context, lack of musical skills later in life is puzzling.

Musical deficits are particularly intriguing when they emerge in isolation from speech delay, intellectual disability, acquired brain damage, or music deprivation. These musical deficits are indicative of congenital amusia. The most common form of congenital amusia concerns the processing of the pitch structure of music and is often referred to as pitch deafness. Individuals with pitch deafness have a normal understanding

* Corresponding author. BRAMS - Pavillon Marie-Victorin, FAS - Département de psychologie, CP 6128, succ. Centre-Ville, Montréal, H3C 3J7 QC, Canada.

E-mail addresses: marie-elaine.lagrois@umontreal.ca (M.-É. Lagrois), isabelle.peretz@umontreal.ca (I. Peretz).

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of speech and prosody in everyday life. They can recognize speakers by their voices and can identify all types of familiar environmental sounds, such as animal cries. What characterizes them behaviorally is their difficulty with detecting out-of-tune singing, including their own, recognizing a familiar tune without the aid of the lyrics, discriminating melodies varying in pitch, and maintaining such melodies in short-term memory (e.g., Ayotte, Peretz, & Hyde, 2002).

Major progress has been made in recent research with regard to the neurobiological etiology of this musical pitch disorder (Peretz, 2016). Pitch deafness is marked by a neural anomaly affecting functional and structural connectivity between the right auditory cortex and inferior frontal cortex. It is also hereditary. Thus, congenital amusia represents a rare chance to examine the neurobiology of music cognition by tracing causal links between genes, brain, and behavior. The logic is essentially one of reverse engineering. An anomaly observed at the behavioral level can be traced back to cognitive processes, then to neurophysiological processes, and ultimately to genes and environment. Accordingly, the identification of associated behavioral deficits is essential. Here, we examine to what extent the pitch deficit characterizing pitch deafness is related to a deficit in abstracting properties of temporal structure from music, namely its beat.

Deficits in beat processing, initially called beat deafness (Phillips-Silver et al., 2011), can occur in isolation (Béglé et al., 2017; Dalla Bella & Sowiński, 2015; Phillips-Silver et al., 2011; Sowiński & Dalla Bella, 2013; Tranchant, Vuvan, & Peretz, 2016), but may also occur in association with pitch deficits. About half of individuals with pitch deafness also show impairments on tasks requiring rhythm discrimination (Ayotte et al., 2002; Peretz, Champod, & Hyde, 2003). Previous studies have shown that, in pitch deafness, the presence of pitch variations interferes with the detection of a temporal change in sound sequences (Foxton, Nandy, & Griffiths, 2006; Hyde & Peretz, 2004; Pfeuty & Peretz, 2010). The available research suggests that when pitch variations are removed, discrimination of rhythmic patterns returns to normal (Foxton et al., 2006; Phillips-Silver, Toiviainen, Gosselin, & Peretz, 2013). These findings have led to the conclusion that the rhythmic deficit found in pitch deafness is a cascade effect of inadequate processing of musical pitch (Dalla Bella & Peretz, 2003; Hyde & Peretz, 2004; Pfeuty & Peretz, 2010).

This “pitch interference account” of the associated rhythm deficit in pitch deafness has limitations. If it was the case that rhythm processing is compromised by a faulty pitch processing system, then all individuals with pitch deafness should show a musical rhythm deficit to some extent. As mentioned above, a rhythmic problem does not occur in all cases, but in about half of sampled amusics. Similarly, one would expect to find a correlation between the severity of the pitch impairment and the severity of the associated rhythmic deficit. Foxton et al. (2006) looked at the possible association between perception of pitch intervals and time intervals in pitch-deaf amusics and found no such correlation. This suggests that the pitch and time deficits may be distinct in congenital amusia. Here, we re-examine the co-occurrence of a rhythm deficit in pitch deafness with natural

musical stimuli, where pitch variations are embedded (Experiment 1) or reduced (Experiment 2).

In Experiment 1, we tested beat perception and synchronization to natural music using an adaptation of the Beat Alignment Test (BAT, Iversen & Patel, 2008). In this test, participants tap to the beat of the musical stimuli (production task) and also judge whether a superimposed metronome track is aligned with the beat of the same stimuli (perception task). About half of the individuals with pitch deafness were expected to perform poorly in these beat perception and production tasks. If rhythm and pitch deficits are distinct in congenital amusia, the beat finding disorder should be unrelated to the severity of the pitch deficit. In order to test these predictions more directly, in Experiment 2, beat finding abilities were assessed in the same participants with drum versions of a subset of the stimuli used in Experiment 1.

2. Experiment 1: beat alignment tests in natural music

2.1. Method

2.1.1. Participants

Ten participants who met the diagnostic criteria for the pitch-deaf form of congenital amusia (age: 43.6 ± 18.0 years; eight females) and a matched control group of 12 participants (age: 42.4 ± 18.2 years; nine females) took part in the study. Controls were further matched to the pitch-deaf group in education and years of music and dance training. Detailed group characteristics are provided in Table 1. Participants provided written consent and received monetary compensation for their participation. All procedures were approved by the Research Ethics Council for the Faculty of Arts and Sciences at the Université de Montréal.

Prior to being selected for participation in this study, the participants underwent tests of their musical abilities. Pitch-deaf participants were included in this study based on their scores on both the online test (Peretz & Vuvan, 2017) and the Montreal Battery for Evaluation of Amusia (MBEA, Peretz et al., 2003). The online test is composed of three tests: Scale, Off-beat, and Off-key. The Scale test is the same in the online test and in the MBEA; it involves the comparison of 30 pairs of melodies that differ by an out-of-key note in half of the trials. The Off-beat and Off-key tasks require the detection of an out-of-time and out-of-key note in a melody, respectively. All control participants had scores within 2 SDs of the mean, indicating normal music perception. The MBEA comprises five additional tests: Contour, Interval, Rhythm, Meter and Memory. The score on the first two tests and the Scale test can be averaged in a melodic composite score, which gives an indication of participants' ability to detect pitch deviations in a melodic context. A score lying 2 SDs below the mean of control groups for the Melodic Composite score of the MBEA, or for both scores on the Scale and Off-key subtests of the online test, indicates the presence of pitch deafness (Peretz & Vuvan, 2017; Vuvan et al., 2017; Table 1).

Table 1 – Characteristics of amusic and matched control participants.

Characteristic	A1	A2	A3	A4	A5	A6	A7	A8	A9	A10	Control Group (Range)
Gender	F	F	F	F	M	M	F	F	F	F	9F/3M
Age (years)	60	25	32	55	31	30	59	58	18	68	42.4 (23–72)
Education (years)	19	16	19	19	19	21	20	15	14	18	17.1 (12–25)
Music Training (years)	2	1	0	5	0	0	0	0	0	1	.5 (0–3)
Dance Training (years)	0	0	0	0	0	0	0	0	0	1	.7 (0–6)
Online Test^a											
Scale (22/30)	20	15	19	23	21	22	14	22	22	20	27 (22–29)
Off-beat (17/24)	19	15	18	20	17	19	18	17	19	18	20 (17–21)
Off-key (16/24)	19	12	13	15	16	13	16	9	14	15	20 (17–22)
MBEA^a											
Melodic Composite (21.4/30)	16.3	17.8	18	20	20.3	20.3	21	22*	22*	22.7*	–
Rhythm (22/30)	22	18	25	25	18	22	22	25	24	22	–
Meter (17/30)	20	25	22	25	20	15	16	26	27	22	–
25 cents pitch-change detection (% accuracy)	30.0	21.1	63.3	53.3	N/A	81.7	10.4	40.6	77.8	57.8	92.1 (75.6–100) ^b

Note: M = male; F = female; MBEA = Montreal Battery of Evaluation of Amusia. ^a Scores in parentheses indicate the cut-off score for each test from Peretz and Vuvan (2017, online test) and Vuvan et al. (2017, MBEA). Score in bold indicates a deficit. * Below cut-off according to earlier norms (Peretz et al., 2003). ^b From an additional control group (n = 30, mean age: 52.5 years old).

The Rhythm and Meter tests of the MBEA reflect different aspects of musical rhythm processing. The Rhythm test consists of comparing pairs of melodies where the temporal grouping in the comparison melody differs in half the trials. The Meter test consists of judging if a melody is a march or a waltz. As can be seen in Table 1, two pitch-deaf participants (A1 and A10) had scores below the cut-off on the Rhythm test and two others (A5 and A9) had scores below the cut-off on the Meter test. Thus, four of the 10 pitch-deaf amusics show indications of a rhythm problem in processing music using these tasks as typically observed in previous studies.

In order to get an index of the severity of the pitch deficit experienced by pitch-deaf amusics, they were tested on a pitch-change detection task. In this task, participants hear sequences of five tones and are asked to detect whether the fourth tone changes in pitch. This task is performed as part of the protocol for identification of pitch-deaf individuals in our research group (e.g., Vuvan et al., 2017). Here, we report detection accuracy for pitch changes of a quarter semitone (25 cents), the smallest pitch change included in the task, which is the most discriminant in comparison to neurotypical adults (Hyde & Peretz, 2004; Vuvan, Nunes-Silva, & Peretz, 2015).

All pitch-deaf participants had normal non-verbal reasoning and verbal working memory abilities as assessed by the Matrix Reasoning and the Digit Span tests from the WAIS-III (Wechsler Adult Intelligence Scale; Wechsler, Coalson, & Raiford, 1997).

2.1.2. Materials and procedure

The Montreal version of the Beat Alignment Test (M-BAT, Tranchant, Lagrois, Bellemare Pépin, G.Schultz, & Peretz, 2018; BAT, Iversen & Patel, 2008) includes a beat tapping task and a beat perception task. In both tasks, our version of the BAT presented the same ten musical excerpts of pop and jazz music at various tempi (range: 82–170 beats per minute). The music excerpts lasted between 23 and 31 sec and contained at least 24 beats.

In the beat production task, participants were asked to tap along to the beat of the musical stimuli. The 10 excerpts were presented twice, in two distinct blocks, for a total of 20 trials. When the concept of beat was not clear to the participant, it was described as the “tic-toc” of a clock. Participants received four practice trials on musical excerpts that were not part of the test. After each practice trial, the music was presented with a click track superimposed on the beat to make it clear where taps were expected. The presentation order of the stimuli was randomized for each participant. The beat tapping task was always performed first to control for exposure to clicks on the beats of the stimuli in the perception task.

Isochronous clicks were superimposed on the music track for the beat perception task. On half of the trials, the clicks were “on beat” and on the other half “off-beat” by either a phase shift ($\pm 15\%$) or a period shift ($\pm 5\%$). The click series started five seconds after each excerpt commenced playing and always included 24 clicks. The presentation order was pseudo-randomized so that no song was presented twice consecutively. The task included 80 trials (eight repetitions of the ten musical excerpts). Participants judged at the end of each stimulus if the clicks were on the beat or not, using four response choices presented on screen: *always on the beat* (1), *mostly on the beat* (2), *sometimes on the beat* (3) and *rarely or never on the beat* (4). For the analyses, the first two choices were considered “on beat” responses and the last two “off-beat” responses. Before starting the task, participants received six practice trials with feedback on accuracy.

The experiment took place in a large sound-attenuated studio. The stimuli were delivered through headphones (DT 770 PRO, Beyerdynamic) at a comfortable level. The tapping test was programmed with MAX-MSP (<https://cycling74.com>) and the perception test was programmed with MATLAB (<https://www.mathworks.com>). The taps were recorded on a square force sensitive resistor (3.81 cm, Interlink FSR 406) connected to an Arduino Duemilanove microcontroller board (arduino.cc) running an adapted Tap Arduino script (based on

fsr_silence_cont.ino; Schultz & van Vugt, 2016; van Vugt & Schultz, 2015, p. 16178) to transmit timing information to a PC (HP ProDesk 600 G1, Windows 7) via the serial USB port.

2.1.3. Data analysis

A measure of sensitivity (d') of discrimination between “on-beat” and “off-beat” trials was considered for the beat perception test. Correct detection of “off-beat” trials were counted as hits while “off-beat” responses to an “on-beat” trial were considered a false alarm.

For the beat production task, taps were first pre-processed to remove inter-tap intervals (ITIs) that were more than half smaller or larger than the individual median ITI produced (median ITI \pm [median ITI*0.5]). This resulted in one to nine taps per trial being removed. Trials with fewer than eight taps were discarded because the analysis of synchronization is more prone to bias with a small number of data points. However, the number of trials eliminated was low, with at least 18 out of the 20 trials being analyzable for each participant. In order to analyze performance on the same beats across the beat perception and beat production tasks, taps produced during the first ten and last five seconds of each song were discarded. Thus, 24 beats of each song were considered for analysis.

Synchronization with the song beat was measured with circular statistics using the Circular Statistics Toolbox for MATLAB (Berens, 2009). With this technique, taps are transposed as angles on a circle from 0 to 360°, where a full circle corresponds to the inter-beat interval. The position of the taps on the circle is used to calculate a mean resultant vector. The length of the mean resultant vector indicates how clustered are the points around the circle. Vector length (VL) ranges from 0 to 1; the larger the value, the more clustered together are the points on the circle, indicating that the period (or time interval) between taps tends to match the inter-beat interval of the stimulus more consistently (see Dalla Bella & Sowiński, 2015, where the same procedure was used). Statistical analyses performed on vector length used a logit transform ($\log VL = -1 \cdot \log [1 - VL]$) because vector length distribution is typically skewed in synchronization data. For simplicity, untransformed vector length is reported when considering group means and individual data. The Rayleigh z test of periodicity was further used to test if participants' taps had a consistent relationship with the inter-beat period, thus indicating if participants could match the period of the beat with their taps (Wilkie, 1983). A significant Rayleigh test (p -value $< .05$) indicates successful period matching between taps and the inter-beat interval of the stimulus. Trials with a p -value $< .05$ on the Rayleigh were thus considered as trials with successful period matching; the percentage of trials with successful period matching was computed for each participant. The inter-beat interval used to generate the mean resultant vector and to perform the Rayleigh test was adjusted to fit the metric level (beat period) at which participants tapped on each trial. Three beat periods were considered for each stimulus: the beat period corresponding to the tempo of the song, half the beat period of the tempo, and twice the beat period of the tempo. Based on the mean ITI of a participant for a given song, the closest beat period from that song was chosen to compute circular statistics.

We also computed the coefficient of variation (CV = SD ITI / Mean ITI), which is a standard measure of the regularity of the ITI that does not take into account the period of the stimuli. The smaller the CV, the less variable are the tap intervals.

2.2. Results and comments

2.2.1. Beat perception

The average percentage of hits minus false alarms was 72.5% for controls (range: 50.0%–97.5%) and 26.8% for pitch-deaf participants (range: 10.0%–60.0%). The derived d' indices were significantly different between the two groups, $t(20) = 4.40$, $p < .001$. Nevertheless, three of the 10 pitch-deaf (A2, A4, A9) participants performed within the controls' range (Fig. 1A). Note that each control participant obtained a normal score according to our norms (Tranchant et al., 2018).

2.2.2. Beat production

On average, the control group successfully matched their taps to the inter-beat interval (IBI) of the songs on 96.6% of trials (range: 85%–100%; Fig. 1B), whereas most pitch-deaf participants were quite poor at matching their taps to the IBI of the song ($M = 57.2\%$ of trials; range: 20%–95%). Still, four pitch-deaf participants (A2, A4, A7, A9) could match the period of their taps to the beat of most songs. Three of them (A2, A4, A9) also performed on par with control participants for this beat perception task (see Fig. 1). No control showed impairment in that task.

The mean vector length (VL) in control participants was .90 ($SD = .04$). The average vector length in the pitch-deaf group was .53 ($SD = .27$, range: .22–.87) and differed significantly from the control group, $t(14.5) = 5.6$, $p < .001$ (comparison with $\log VL$; Table 2). Two of the ten pitch-deaf amusics had vector length similar to controls (A4: VL = .87, and A9: VL = .86).

The mean CV of the control group was .07 ($SD = .01$), while the mean CV for pitch-deaf group was .10 ($SD = .03$, range: .08–.16). The mean CV differed significantly between groups, $t(20) = -3.86$, $p = .001$. Thus, all but two pitch-deaf individuals (A4 and A9) tapped less regularly and were less consistently aligned with the period of the stimuli than controls (Table 2).

2.2.3. Relation between pitch and beat deficits

In order to assess a possible relationship between pitch and beat processing abilities, we computed the Pearson correlation coefficient between the scores obtained on the Scale test of the online test (as all participants had completed it) and the d' scores obtained in the perception task (Fig. 2A). The scores were highly correlated, $r_{(20)} = .68$, $p = .001$. This was also the case for the mean $\log VL$, with $r_{(20)} = .64$, $p < .001$ (Fig. 2B). Interestingly, the percentage of accurate pitch-change detection in acoustical sequences (Table 1) did not predict pitch-deaf participant's performance on the beat perception and production tasks, with $r_{(7)} = -.20$, $p = .61$ and $r_{(7)} = -.17$, $p = .67$ for d' and $\log VL$, respectively, using Spearman non-parametric correlation coefficient. This is illustrated by the observation that A2, A4, A7, and A9, who were quasi-normal at tracking the beat of music for synchronization purposes, were impaired in the pitch-change detection task (Table 1). The term “quasi-normal” reflects the fact that A2 and A4 were less consistent than control participants in synchronization and A7 was impaired in beat perception.

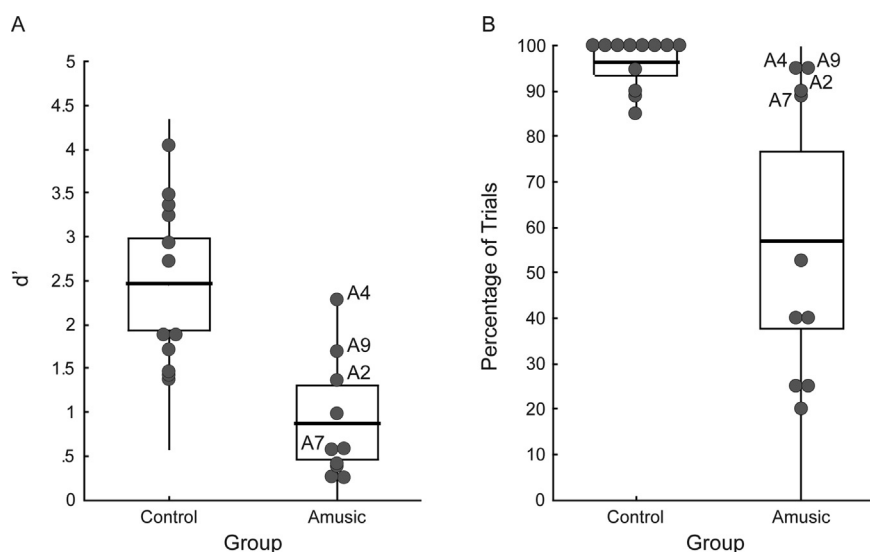


Fig. 1 – Participants' performance on the M-BAT. A. d' scores on the beat perception task of the M-BAT. Error bars represent two standard deviations from the mean. B. Percentage of trials with successful period matching on the beat production task of the M-BAT. Each dot represents a participant.

Table 2 – Mean vector length (VL) and coefficient of variation (CV) in the M-BAT production task.

	A1	A2	A3	A4	A5	A6	A7	A8	A9	A10	Control Group Mean (Range)
VL	.50	.78	.29	.87	.22	.40	.80	.25	.86	.35	.90 (.83–.95)
CV	.10	.10	.11	.09	.14	.09	.16	.09	.08	.10	.07 (.06–.09)

Note: Amusic participants in bold were comparable to controls for both the VL and the CV.

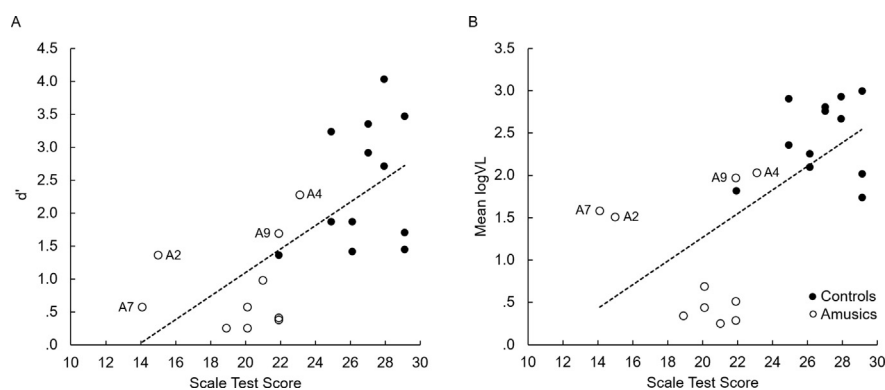


Fig. 2 – Illustration of Correlation Between the Scale Test Score from the Online Test of Amusia and Performance in the M-BAT. A. Correlation between the Scale test score and d' on the beat perception task. B. Correlation between the Scale score and the mean logVL on the beat production task. Controls are marked by black dots and pitch-deaf amusics by white dots. Pitch-deaf participants A2, A4, and A9 performed like controls on the M-BAT.

3. Experiment 2: synchronization to drum rhythms

The co-occurrence of the pitch deficit with a beat deficit revealed in Experiment 1 in the majority of the pitch-deaf amusics called for a re-examination of beat finding performance in a context where their pitch deficit was unlikely to

interfere with beat finding abilities, in case the latter was intact. This was tested in Experiment 2 with percussive music.

3.1. Method

The same participants were tested with percussive renditions of *Suavemente* (by Elvis Crespo), played at a tempo of 112 bpm and 120 bpm, with the audio files lasting 36 sec and 33 sec,

respectively. This procedure has been used previously with a different pool of participants (Phillips-Silver et al., 2013). Each version of the song contained 65 beats that were created with a snare drum, a tenor drum, and a bass drum, so as to reproduce as closely as possible the major instrumental lines of the original song (for a detailed description of these stimuli, see Phillips-Silver et al., 2013). We added a percussive rendition of the song *Brand New Carpet* (by Bodi Bill), similarly created, at 126 bpm with a duration of 16 sec. This stimulus had 31 beats. Presentation order of the excerpts was counterbalanced, with *Brand New Carpet* always played in between the two drum versions of *Suavemente*. The original versions of *Suavemente* and *Brand New Carpet* were presented as stimuli in Experiment 1 and could therefore serve here for comparison.

Participants were asked to tap to the beat of the stimuli. A practice trial was performed before starting the task to make sure they understood the instructions. The practice trial used a drum rhythm not included in the test. In addition, the participants were asked to synchronize their taps to a metronome. This control task was included to assess sensorimotor synchronization when there was no need for beat extraction. Participants listened to seven metronome ticks and then had to synchronize their taps to a metronome at the same tempo for 60 taps. The task comprised two trials, one at a tempo of 96 bpm and one at 120 bpm. Each metronome stimulus was composed of 440 Hz sine wave ticks, each with a duration of 50 msec. The presentation order of the two metronome stimuli was counter-balanced between participants. A practice trial for metronome synchronization at 108 bpm was presented first to make sure participants understood the instructions.

Taps were recorded with the same system described in Experiment 1 section 2.1.2, with the stimuli again presented through headphones.

Circular statistics were used to assess synchronization as described in Experiment 1 section 2.1.3. In order to remove initial variability in synchronization, the first five beats of each drum excerpt were discarded from the analysis; the next 24 beats were considered for the analysis to allow comparison with the results from the production task of the M-BAT. For the synchronization to the metronome, the first five beats were discarded to remove initial variability, leaving 55 beats for analysis.

3.2. Results and comments

3.2.1. Results of the tapping task

All but one control participants successfully matched their taps to the period of the three drum trials and so did four of the ten pitch-deaf participants (A2, A4, A7, and A9). The one

control participant who failed to synchronize to *Brand New Carpet* ($p = .72$) was able to synchronize to both *Suavemente* trials. In contrast, one pitch-deaf (A3) participant could only match his taps with the beat period of *Brand New Carpet*.

The four pitch-deaf participants who could synchronize to all drum trials also succeeded in synchronizing their taps to the beats of both trials of the original songs in Experiment 1 (Table 3). These four “beat-preserved” pitch-deaf individuals could also anticipate the beat with a mean negative asynchrony between taps and beats ($M = -39$ msec, $SD = 30$ msec). Controls mean asynchrony was -14 msec ($SD = 22$ msec). These results indicate that the “beat-preserved” pitch-deaf participants showed a similar phase relationship with the beat to that shown by controls.

Tapping performance obtained here, with the drum rhythms, was compared to the performance obtained with the original versions containing pitch variations (Experiment 1) by looking at the mean vector length (VL) and tapping variability (CV) (Table 4). An ANOVA performed on the mean logVL with Group as the between-subjects variable and Condition (drum vs original) as a within-subject variable revealed a main effect of Group, $F(1,20) = 19.03$, $p < .001$, $\eta^2 = .49$, a main effect of Condition, $F(1,20) = 4.81$, $p = .04$, $\eta^2 = .19$, and no significant Group \times Condition interaction, $F(1,20) = .62$, $p = .44$. The group of pitch-deaf participants obtained a smaller VL (.50) than controls (.91) overall. Both groups had smaller VL with the drum versions than the original songs, although the effect was more salient in the pitch-deaf group (Table 4). The mean CVs showed similar trends. These results show that contrary to expectations, pitch-deaf participants synchronized their taps better to the original songs that included pitch variations than to the drum versions. With the latter, the majority of pitch-deaf amusics showed evidence of a beat deficit. The correlation between the mean VL obtained for each version was almost significant in the pitch-deaf group, with $r_{(8)} = -.62$, $p = .054$, and clearly significant in controls, $r_{(10)} = .65$, $p = .02$, using Spearman's correlation for nonparametric data.

All control and all but one (A8) pitch-deaf participants could successfully synchronize their taps to the period of the metronome at both 120 bpm and 96 bpm (Rayleigh test, $p < .05$). A8 successfully synchronized his taps to the metronome at 96 bpm only. Synchronization at 120 bpm was inaccurate because this participant tapped too fast (mean ITI of 427 msec) relative to the 500 msec period of the stimulus. Comparing the length of the resultant vector (logVL) for participants with successful synchronization, we found no significant difference between pitch-deaf and control participants, $F(1,19) = 3.27$, $p = .09$, no effect of Tempo, $F(1,19) = .003$, $p = .96$, and no Group \times Tempo interaction, $F(1,19) = .33$, $p = .57$. Similarly, for the mean asynchrony

Table 3 – Number of trials with successful period matching in the drum and original versions of the songs in the amusic group.

Version	A1	A2	A3	A4	A5	A6	A7	A8	A9	A10
Drums	0/3	3/3	1/3	3/3	0/3	0/3	3/3	0/3	3/3	0/3
Original	2/4	4/4	1/4	4/4	2/4	2/4	4/4	1/4	4/4	2/4

Note: The four “beat-preserved” participants with pitch deafness are indicated in bold.

Table 4 – Individual Pitch-deaf Participants' Mean Vector Length (VL) and Mean Coefficient of Variation (CV) of Tapping Performance to the Drum and Original Versions of the Songs. Group values for controls are included for comparison.

	VL		CV	
	Drums	Original	Drums	Original
A1	.09	.59	.15	.10
A2	.66	.79	.12	.14
A3	.20	.22	.09	.13
A4	.87	.96	.07	.05
A5	.10	.27	.14	.12
A6	.17	.58	.09	.08
A7	.95	.93	.10	.15
A8	.23	.21	.12	.10
A9	.81	.90	.08	.10
A10	.08	.31	.09	.09
Control Group Mean (Range)	.89 (.67–.98)	.92 (.77–.97)	.06 (.04–.10)	.07 (.05–.13)

Note: Normal performance in pitch-deaf participants is in bold.

between the taps and the onsets of the metronome beat, there was no main effect of Group, $F(1,19) = 1.67$, $p = .21$, no effect of Tempo, $F(1,19) = .30$, $p = .59$, and no significant interaction, $F(1,19) = 1.28$, $p = .27$. The two groups showed mean negative asynchronies to both tempi: controls' $M = -48$ msec (range: -115 msec– 4 msec), pitch-deaf amusics' $M = -66$ msec (range: -161 msec to -14 msec). Thus, as shown in previous studies, pitch-deaf amusics could synchronize to the metronome as accurately as controls, suggesting no general sensorimotor synchronization deficits (Dalla Bella & Peretz, 2003; Phillips-Silver et al., 2013).

3.2.2. Relation between tapping to drums and musical pitch processing

As in Experiment 1, the correlation between the scores obtained on the Scale test and the mean logVLs obtained for

drum rhythms in this experiment was significant, $r_{(20)} = .46$, $p = .03$ (Fig. 3). This is despite the presence of clear outliers among the pitch-deaf group (A2, A4, A7, A9), who displayed normal synchronization with the drums' beat and poor musical pitch perception.

4. General discussion

The main finding of the present study is that melody and beat impairments are associated in most cases of pitch deafness. In our sample of ten adults diagnosed as having a deficit in musical pitch processing, at least six also manifest a deficit in finding the musical beat in music and drum rhythms. However, the presence of two to four clear-cut cases of musical pitch disorder with spared beat processing abilities suggests that the pitch and beat deficits are distinct disorders. In what follows, we discuss the possible origins of the frequent co-occurrence of the musical deficits and the implications for the behavioral characterization of congenital amusia.

The attribution of the rhythmic difficulties to the possible interference caused by inadequate processing of pitch variations (i.e., the pitch interference hypothesis) finds little support in the present study. The beat finding deficit experienced by the majority of pitch-deaf amusics remains severe whether pitch cues are present or not in the musical stimulus. Moreover, the beat-impaired amusics better align their taps to the original music, which contains pitch variations, than to their percussive renditions, although matched control participants do not show such a clear advantage for the original music. Thus, the presence of putative interfering pitch information does not appear to play a significant role in the occurrence of the beat deficit.

Yet, there is a correlation between the severity of the musical pitch disorder and the size of the beat deficit, especially in perception (Fig. 2A). This relation holds for amusics and controls alike. The higher the score in discriminating melodies, in which there can be a changed note that violates the melodic pitch structure (on the Scale test of the MBEA), the higher the detection of misalignments of metronome clicks superimposed on music (on the M-BAT test). Obviously, the observed correlation between pitch and beat performance

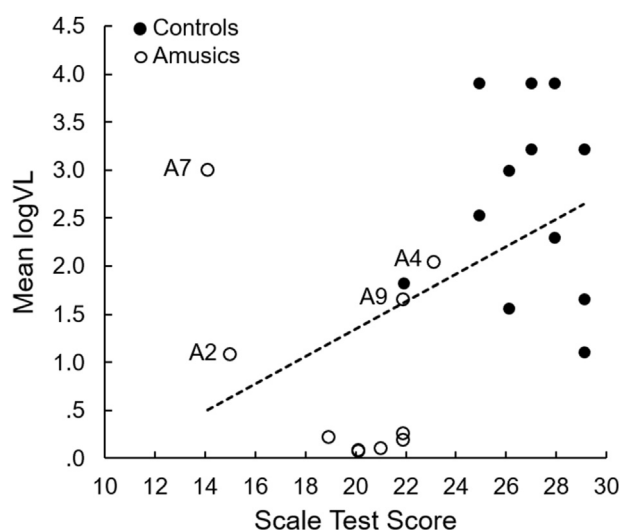


Fig. 3 – Illustration of the Correlation Between the Scale Test Score and Mean LogVL When Tapping to Drum Rhythms. Control participants are marked with black dots and pitch-deaf amusic participants with white dots. Marked pitch-deaf participants A2, A4, A7, and A9 exhibited normal synchronization with the drums' beat.

could be due to several factors that are not specific to music structure, such as auditory attention and motivation. Nevertheless, given the presence of correlations across tests of pitch and beat processing and the frequent co-occurrence of deficits in the processing of the two, the possibility of shared processing components should be examined.

Shared mechanisms between pitch and beat processing could occur at several levels, from sensory input through to motor output. Here, we can discard the two end processes since the basic auditory-motor loop appears normal in pitch-deaf amusics. First, there was no correlation between the severity of the sensory impairment observed in pitch-change detection in five-tone isochronous sequences and the tested beat finding abilities, suggesting no direct association between acoustic pitch and beat processing. Secondly, all ten pitch-deaf individuals were able to accurately match their taps to auditory metronome sequences, suggesting intact basic auditory-motor coupling in the context of a tapping task. Thus, shared mechanisms between pitch and beat processing are likely to concern more cognitive components. There is substantial evidence for interaction between pitch and time dimensions in music, although these are separable processing components. For example, a mismatch between pitch and temporal accents, or an atonal melodic context, can lower the capacity to track beats (Ellis & Jones, 2009; Jones & Pfordresher, 1997; Pfordresher, 2003; Prince, 2011, 2014; Prince & Pfordresher, 2012). The question of how information from pitch and time combines in music has been an area of continued interest (see Krumhansl, 2000; Prince, 2011, for reviews), with unfortunately little consensus on the issue of whether the integration of these dimensions is additive (Palmer & Krumhansl, 1987a; 1987b) or interactive (Jones, 1987; Jones & Boltz, 1989) and at what stage in the decision process the two dimensions are integrated. Hence, the identification of a shared processing component will have to await future development of cognitive models.

Identification of the locus for the observed tight association underlying pitch and beat processing might be aided by knowing their neural correlates. Here again, current knowledge gained from neuroimaging studies is not very informative or sufficiently constraining to provide good candidates for shared processing components. In a recent study (Sihvonen et al., 2016), both musical pitch and rhythm processing were examined in 77 brain-damaged patients while using the same screening tests used here, namely the Scale and the Rhythm tests of the MBEA. Deficits in each test were associated with lesions in the auditory cortex, Heschl's gyri, insula, and basal ganglia (putamen, caudate, pallidum) of the right hemisphere. Thus, a common locus for processing both types of structure may lie in that constellation of regions. However, we saw here that our pitch-deaf amusics with a beat finding disorder had normal scores on the rhythm test of the MBEA. Moreover, Grahn and McAuley (2009) found that good beat finders have greater brain activity in the supplementary motor area, left premotor cortex, and left insula, while poor beat-perceivers show relatively greater activation in the left posterior superior and middle temporal gyri and the right premotor cortex. These brain regions do not overlap with the anomalous fronto-temporal network identified in pitch deafness. Pitch deficits in congenital amusia have been linked to anomalies in

connectivity between the inferior frontal gyrus (IFG; BA 44/45/47) and the superior temporal gyrus (STG; BA 22). More precisely, deficient connections from the right IFG to the right STG would prevent top-down influence from higher-order cortical regions in pitch processing (for a recent review see Peretz, 2016). Therefore, there is at present no clear indication of how or where in the brain the pitch and beat defects might overlap.

Nevertheless, there is a need to identify the co-occurrence of pitch and time deficits in congenital amusia in order to progress the characterization of the disorder. While there is a large consensus on how to screen for the presence of musical pitch deficits by using, among other tests, the Scale test of the MBEA (Vuvan et al., 2017), there is no equivalent consensus for beat deficits. Here we show that none of the MBEA tests is appropriate, not even the MBEA meter test that is supposed to tap the beat finding abilities. Yet, Phillips-Silver et al. (2013) found a positive correlation between the scores on the Meter test of the MBEA and beat synchronization with the same *Suavemente* song used here for the evaluation of beat finding abilities. We do not corroborate this finding since none of the correlations between the MBEA meter test and the synchronization measures considered here reached significance. The reasons for this discrepancy between the prior and current studies are unclear. Therefore, in future studies, we propose to use the M-BAT test for its sensitivity to the presence of a beat deficit (see also Tranchant et al., 2018, for norms on this test) rather than the MBEA meter test. However, the BAT requires the recording of precise motor responses aligned with a stimulus, which can hardly be done outside the lab. One future alternative tool is the BAASTA: Battery for the Assessment of Auditory Sensorimotor and Timing Abilities, which is currently being developed for the tablet using a touch screen (Dalla Bella et al., 2017; Puyjarinet, Bégel, Lopez, Dellacherie, & Dalla Bella, 2017).

Another area of research that would deserve more attention regarding congenital amusia is whether this population could benefit from musical intervention to improve performance. A few prior studies have been conducted to test if pitch perception could be improved in pitch deafness and results have so far been mostly negative (e.g., Hyde & Peretz, 2004; Liu, Jiang, Francart, Chan, & Wong, 2017; Mignault Goulet, Moreau, Robitaille, & Peretz, 2012). One recent study (Whiteford & Oxenham, 2018) obtained promising results after only five training sessions of pitch-change detection, although the contribution of a practice effect from test-retest could not be excluded since pitch-deaf participants trained on an irrelevant task also improved from pre-test to post-test. So far, training of beat processing abilities has not yet been assessed in amusics. Phillips-Silver et al. (2013) noted that in their group of pitch-deaf participants the accuracy of synchronization to the beat, when bouncing to a musical excerpt, tended to improve between the first and second trial. A follow-up with one of the pitch-deaf participants also showed an improvement in synchronization performance a few months later. In our study, we could not assess practice effect on beat finding abilities since presentation order was randomized for each participant. However, in the synchronization task of the M-BAT, which consists of the repetition of the same songs in successive blocks, we did not find an increase in performance.

Future studies should examine more closely the distinct effect of practice and intervention in congenital amusia.

A promising strategy for training rhythmic skills, called *Rhythm Workers*, has recently been developed (Bégel, Seilles, & Dalla Bella, 2018). The training consists of a beat production (tapping) task or a beat perception task, both implemented on a tablet, using musical excerpts of various beat complexity. The tasks used in the training protocol and to measure pre-post change in performance are very similar to the M-BAT used here. Preliminary testing of the protocol indicates improvement in beat perception assessed before and after training in young neurotypical adults over a two weeks home-based training period (Bégel et al., 2018) as well as in patients with Parkinson's disease, over a six weeks training period (Dauvergne et al., 2018). Transfer of improvements to different movements than tapping and beat perception in general remains to be addressed.

In summary, we have shown that pitch and time deficits more often co-occur in congenital amusia than they dissociate. This finding highlights the tight connection between melody and rhythm in music processing and invites researchers to systematically test for the joint presence of these deficits to contribute to the understanding of the origins of these neurodevelopmental disorders that are presently considered distinct.

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Disclosure statement

The authors declare no competing interests. No part of the study procedures nor part of the study analyses were pre-registered prior to the research being conducted.

Open practices

The study in this article earned an Open Data badge for transparent practices. Materials and data for the study are available at <https://doi.org/10.6084/m9.figshare.7271480.v1>.

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Supplementary data

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REFERENCES

- Ayotte, J., Peretz, I., & Hyde, K. (2002). Congenital amusia: A group study of adults afflicted with a music-specific disorder. *Brain*, 125(2), 238–251. <https://doi.org/10.1093/brain/awf028>.
- Bégel, V., Benoit, C.-E., Correa, A., Cutanda, D., Kotz, S. A., & Dalla Bella, S. (2017). “Lost in time” but still moving to the beat. *Neuropsychologia*, 94, 129–138. <https://doi.org/10.1016/j.neuropsychologia.2016.11.022>.
- Bégel, V., Seilles, A., & Dalla Bella, S. (2018). *Rhythm Workers: A music-based serious game for training rhythm skills*. *Music & Science*, 1. <https://doi.org/10.1177/2059204318794369>.
- Berens, P. (2009). *CircStat: A MATLAB toolbox for circular statistics*. *Journal of Statistical Software*, 31(10), 1–21.
- Cirelli, L. K., Einarson, K. M., & Trainor, L. J. (2014). Interpersonal synchrony increases prosocial behavior in infants. *Developmental Science*, 17(6), 1003–1011. <https://doi.org/10.1111/desc.12193>.
- Dalla Bella, S., Farrugia, N., Benoit, C.-E., Bégel, V., Verga, L., Harding, E., et al. (2017). BAASTA: Battery for the assessment of auditory sensorimotor and timing abilities. *Behavior Research Methods*, 49(3), 1128–1145. <https://doi.org/10.3758/s13428-016-0773-6>.
- Dalla Bella, S., & Peretz, I. (2003). Congenital amusia interferes with the ability to synchronize with music. *Annals of the New York Academy of Sciences*, 999(1), 166–169. <https://doi.org/10.1196/annals.1284.021>.
- Dalla Bella, S., & Sowiński, J. (2015). Uncovering beat deafness: Detecting rhythm disorders with synchronized finger tapping and perceptual timing tasks. *Journal of Visualized Experiments JoVE*, (97), 51761. <https://doi.org/10.3791/51761>.
- Dauvergne, C., Bégel, V., Gény, C., Puyjarinet, F., Laffont, I., & Dalla Bella, S. (2018). Home-based training of rhythmic skills with a serious game in Parkinson's disease: Usability and acceptability. *Annals of Physical and Rehabilitation Medicine*. <https://doi.org/10.1016/j.rehab.2018.08.002>.
- Ellis, R. J., & Jones, M. R. (2009). The role of accent salience and joint accent structure in meter perception. *Journal of Experimental Psychology Human Perception and Performance*, 35(1), 264–280. <https://doi.org/10.1037/a0013482>.
- Foxton, J. M., Nandy, R. K., & Griffiths, T. D. (2006). Rhythm deficits in “tone deafness”. *Brain and Cognition*, 62(1), 24–29. <https://doi.org/10.1016/j.bandc.2006.03.005>.
- Grahn, J. A., & McAuley, J. D. (2009). Neural bases of individual differences in beat perception. *Neuroimage*, 47(4), 1894–1903. <https://doi.org/10.1016/j.neuroimage.2009.04.039>.
- Hyde, K. L., & Peretz, I. (2004). Brains that are out of tune but in time. *Psychological Science*, 15(5), 356–360. <https://doi.org/10.1111/j.0956-7976.2004.00683.x>.
- Iversen, J. R., & Patel, A. D. (2008). The beat alignment test (BAT): Surveying beat processing abilities in the general population. In *Proceedings of the 10th international conference on music perception and cognition (ICMPC10)*, Sapporo.
- Jones, M. R. (1987). Dynamic pattern structure in music: Recent theory and research. *Perception & Psychophysics*, 41(6), 621–634. <https://doi.org/10.3758/bf03210494>.
- Jones, M. R., & Boltz, M. (1989). Dynamic attending and responses to time. *Psychological Review*, 96(3), 459–491. <https://doi.org/10.1037/0033-295X.96.3.459>.
- Jones, M. R., & Pfordresher, P. Q. (1997). Tracking musical patterns using joint accent structure. *Canadian Journal of Experimental Psychology*, 51(4), 271–291. <https://doi.org/10.1037/1196-1961.51.4.271>.
- Krumhansl, C. L. (2000). Rhythm and pitch in music cognition. *Psychological Bulletin*, 126(1), 159–179. <https://doi.org/10.1037/0033-2909.126.1.159>.
- Liu, F., Jiang, C., Francart, T., Chan, A. H. D., & Wong, P. C. M. (2017). Perceptual learning of pitch direction in congenital

- amusia. Evidence from Chinese speakers. *Music Perception: An Interdisciplinary Journal*, 34(3), 335–351. <https://doi.org/10.1525/mp.2017.34.3.335>.
- Mignault Goulet, G., Moreau, P., Robitaille, N., & Peretz, I. (2012). Congenital amusia persists in the developing brain after daily music listening. *Plos One*, 7(5), e36860. <https://doi.org/10.1371/journal.pone.0036860>.
- Palmer, C., & Krumhansl, C. L. (1987a). Independent temporal and pitch structures in determination of musical phrases. *Journal of Experimental Psychology Human Perception and Performance*, 13(1), 116–126. <https://doi.org/10.1037/0096-1523.13.1.116>.
- Palmer, C., & Krumhansl, C. L. (1987b). Pitch and temporal contributions to musical phrase perception: Effects of harmony, performance timing, and familiarity. *Perception & Psychophysics*, 41(6), 505–518. <https://doi.org/10.3758/bf03210485>.
- Perani, D., Saccuman, M. C., Scifo, P., Spada, D., Andreolli, G., Rovelli, R., et al. (2010). Functional specializations for music processing in the newborn human brain. *Proceedings of the National Academy of Sciences*, 107(10), 4758–4763. <https://doi.org/10.1073/pnas.0909074107>.
- Peretz, I. (2016). Neurobiology of congenital amusia. *Trends in Cognitive Sciences*, 20(11), 857–867. <https://doi.org/10.1016/j.tics.2016.09.002>.
- Peretz, I., Champod, A. S., & Hyde, K. L. (2003). Varieties of musical disorders. *Annals of the New York Academy of Sciences*, 999(1), 58–75. <https://doi.org/10.1196/annals.1284.006>.
- Peretz, I., & Vuvan, D. T. (2017). Prevalence of congenital amusia. *European Journal of Human Genetics*, 25, 625. <https://doi.org/10.1038/ejhg.2017.15>.
- Pfeuty, M., & Peretz, I. (2010). Abnormal pitch-time interference in congenital amusia: Evidence from an implicit test. *Attention Perception & Psychophysics*, 72(3), 763–774. <https://doi.org/10.3758/app.72.3.763>.
- Pfordresher, P. Q. (2003). The role of melodic and rhythmic accents in musical structure. *Music Perception: An Interdisciplinary Journal*, 20(4), 431–464. <https://doi.org/10.1525/mp.2003.20.4.431>.
- 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. <https://doi.org/10.1080/02643294.2013.863183>.
- Phillips-Silver, J., Toiviainen, P., Gosselin, N., Piché, O., Nozaradan, S., Palmer, C., et al. (2011). Born to dance but beat deaf: A new form of congenital amusia. *Neuropsychologia*, 49(5), 961–969. <https://doi.org/10.1016/j.neuropsychologia.2011.02.002>.
- Prince, J. B. (2011). The integration of stimulus dimensions in the perception of music. *Quarterly Journal of Experimental Psychology*, 64(11), 2125–2152. <https://doi.org/10.1080/17470218.2011.573080>.
- Prince, J. B. (2014). Pitch structure, but not selective attention, affects accent weightings in metrical grouping. *Journal of Experimental Psychology Human Perception and Performance*, 40(5), 2073–2090. <https://doi.org/10.1037/a0037730>.
- Prince, J. B., & Pfordresher, P. Q. (2012). The role of pitch and temporal diversity in the perception and production of musical sequences. *Acta Psychologica*, 141(2), 184–198. <https://doi.org/10.1016/j.actpsy.2012.07.013>.
- Puyjarinet, F., Bégel, V., Lopez, R., Dellacherie, D., & Dalla Bella, S. (2017). Children and adults with Attention-Deficit/Hyperactivity Disorder cannot move to the beat. *Scientific Reports*, 7(1), 11550. <https://doi.org/10.1038/s41598-017-11295-w>.
- Schultz, B. G., & van Vugt, F. T. (2016). Tap Arduino: An Arduino microcontroller for low-latency auditory feedback in sensorimotor synchronization experiments. *Behavior Research Methods*, 48, 1591–1607. <https://doi.org/10.3758/s13428-015-0671-3>.
- Sihvonen, A. J., Ripollés, P., Leo, V., Rodríguez-Fornells, A., Soinila, S., & Särkämö, T. (2016). Neural basis of acquired amusia and its recovery after stroke. *The Journal of Neuroscience*, 36(34), 8872–8881. <https://doi.org/10.1523/jneurosci.0709-16.2016>.
- Sowiński, J., & Dalla Bella, S. (2013). Poor synchronization to the beat may result from deficient auditory-motor mapping. *Neuropsychologia*, 51(10), 1952–1963. <https://doi.org/10.1016/j.neuropsychologia.2013.06.027>.
- Tranchant, P., Lagrois, M., Bellemare Pépin, A., Schultz, B. G., & Peretz, I. (2018). *Beat alignment test of the motor origin of musical entrainment deficits (Manuscript in preparation)*.
- 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. <https://doi.org/10.1371/journal.pone.0160178>.
- van Vugt, F., & Schultz, B. G. (2015). *Taparduino v1.01*. Zenodo. <https://doi.org/10.5281/zenodo.16178>.
- Vuvan, D. T., Nunes-Silva, M., & Peretz, I. (2015). Meta-analytic evidence for the non-modularity of pitch processing in congenital amusia. *Cortex*, 69, 186–200. <https://doi.org/10.1016/j.cortex.2015.05.002>.
- 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*. <https://doi.org/10.3758/s13428-017-0892-8>.
- Wechsler, D., Coalson, D. L., & Raiford, S. E. (1997). *WAIS-III: Wechsler adult intelligence scale*. San Antonio, TX: Psychological Corporation.
- Whiteford, K. L., & Oxenham, A. J. (2018). Learning for pitch and melody discrimination in congenital amusia. *Cortex*, 103, 164–178. <https://doi.org/10.1016/j.cortex.2018.03.012>.
- Wilkie, D. (1983). Rayleigh test for randomness of circular data. *Applied Statistics*, 32(3), 311–312.
- Winkler, I., Háden, G. P., Ladinig, O., Sziller, I., & Honing, H. (2009). Newborn infants detect the beat in music. *Proceedings of the National Academy of Sciences of the United States of America*, 106(7), 2468–2471. <https://doi.org/10.1073/pnas.0809035106>.
- Zentner, M., & Eerola, T. (2010). Rhythmic engagement with music in infancy. *Proceedings of the National Academy of Sciences*, 107(13), 5768–5773. <https://doi.org/10.1073/pnas.1000121107>.