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Abstract

Congenital amusia is a condition in which an individual suffers from a deficit of musical pitch perception and production. Individuals suffering from congenital amusia generally tend to abstain from musical activities. Here we present the unique case of Tim Falconer, a self-described musicophile who also suffers from congenital amusia. We describe and assess Tim's attempts to train himself out of amusia through a self-imposed 18-month program of formal vocal training and practice. We tested Tim with respect to music perception and vocal production across seven sessions including pre-training and post-training assessments. We also obtained diffusion-weighted images of his brain to assess connectivity between auditory and motor planning areas via the arcuate fasciculus. Tim's behavioural and brain data were compared to that of normal and amusic controls. While Tim showed temporary gains in his singing ability, he did not reach normal levels, and these gains faded when he was not engaged in regular lessons and practice. Tim did show some sustained gains with respect to the perception of musical rhythm and meter. We propose that Tim's lack of improvement in pitch perception and production tasks is due to long-standing and likely irreversible reduction in connectivity along the arcuate fasciculus fiber tract.

Keywords: congenital amusia; diffusion tractography; musical training;

Introduction

Congenital amusia, also known colloquially as tone deafness, is a neurodevelopmental deficit of music perception. It has been proposed that amusia arises from a failure of fine-grained pitch discrimination (Hyde & Peretz, 2004) leading to difficulty in perceiving dissonance (Cousineau, McDermott, & Peretz, 2012; Marin, Thompson, Gingras, & Stewart, 2015) and in making tonal judgments (Peretz, Brattico, Järvenpää, & Tervaniemi, 2009). The specificity of the disorder to pitch perception is reflected in the typical pattern of scores obtained using the Montreal Battery of Evaluation of Amusia (MBEA; Peretz, Champod, & Hyde, 2003). In Peretz, Champod, & Hyde (2003), 23 of 24 amusic participants (amusics hereafter) perform below normal in melody subtests whereas half of them perform as well as controls in the rhythm subtest. Moreover, amusics perform as well as controls on a modified version of the meter subtest, wherein piano pitch variations are replaced with drum sounds (Phillips-Silver, Toiviainen, Gosselin, & Peretz, 2013). Similarly, amusics improve on melody discrimination when stimuli do not vary in pitch (Foxton, Nandy, & Griffiths, 2006). More generally, a recent meta-analysis showed that amusia affects fine-grained processing of pitch in general, not exclusively in music (Vuvan, Nunes-Silva, & Peretz, 2015).

Congenital amusia is diagnosed in individuals independently of their exposure to music. However, this disorder often leads to abstinence from musical experience, which may exacerbate the behavioural and brain differences that are observed between amusics and controls (for a comprehensive review of these neurobiological differences, see Peretz (2016). Although musical training has been associated with structural and functional changes in neurotypical connectivity (Herholz & Zatorre, 2012), similar plasticity has not been reported in amusic individuals so far, neither in the negative direction (i.e., developmental study of the emergence of amusia in

childhood) or in the positive direction (i.e., amelioration of the disorder through music exposure or training; see for instance, Mignault-Goulet et al., 2012). The amusic pitch deficit is associated with anatomical and functional abnormalities along the right fronto-temporal pathway (Hyde, Zatorre, Griffiths, Lerch, & Peretz, 2006; Hyde et al., 2007; Hyde, Zatorre, & Peretz, 2011). This finding is consistent with previous work identifying right auditory cortex and right IFG as primary nodes in the pitch processing network (Zatorre, Evans, & Meyer, 1994). In addition, amusics exhibit diminished functional connectivity within the right fronto-temporal network. Specifically, amusics have stronger functional connectivity between the bilateral auditory cortices and reduced functional connectivity between the right auditory cortex and right IFG in comparison to controls (Albouy et al., 2013; Hyde et al., 2011). Diffusion-weighted imaging further supports this conceptualization, with amusics showing deficient connectivity in the superior right arcuate fasciculus as compared to controls (Loui, Alsop, & Schlaug, 2009), although the robustness of this finding may depend on the analytical algorithms employed (Chen et al., 2015)¹.

The importance of the right arcuate fasciculus in music perception and production is supported by work in neurotypical participants varying in extent and type of musical training. Specifically, Halwani et al. (2011) examined the volume of the arcuate fasciculus in participants with vocal training, instrumental musical training, and no musical training. They found that both groups of musicians differed from non-musicians in that tract volume of the right arcuate fasciculus was larger. When looking at the superior arm of the left arcuate fasciculus, they found that relative to instrumental musicians, singers had a larger tract volume but lower fractional

¹ In fact, Chen et al. (2015) used a different seeding and masking methodology and did not observe any anomaly in the arcuate fasciculus, no matter the analytical algorithm used. This illustrates the importance of methodological features in analyzing functional connectivity.

anisotropy values. Moreover, fractional anisotropy of the superior arm of the left arcuate fasciculus was inversely correlated with the number of years of vocal training. These results suggest that long-term vocal training influences white-matter tracts involved in auditory perception and production, and that these effects can be differentiated from a generalized music training effect.

Although poor pitch perception is usually accompanied by poor pitch production (i.e., singing) in amusics as well as controls (Dalla Bella, Giguère, & Peretz, 2009; Hutchins, Zarate, Zatorre, & Peretz, 2010), a growing body of research suggests that pitch production can dissociate from pitch perception in amusia. For instance, amusics are able to reproduce pitch intervals and adapt to pitch shifts that they have difficulty perceiving (Hutchins & Peretz, 2012; Hutchins & Peretz, 2013; Loui, Guenther, Mathys, & Schlaug, 2008). Moreover, a few amusics appear to be able to sing as well as controls (Dalla Bella et al., 2009; ; Hutchins & Peretz, 2012; Tremblay-Champoux et al., 2010). Importantly, this dissociation indicates the possibility that poor singing in amusia might be more easily improved than the poor pitch perception that is at the core of the disorder.

Previous attempts to train amusics to improve their pitch perception and production have yielded mixed results. In one study, adolescent amusics were exposed to 4 weeks of daily music listening (Mignault Goulet, Moreau, Robitaille, & Peretz, 2012). This musical exposure did not result in any improvement of the amusic participants' pitch thresholds, nor did it change the electrical responses related to the detection of small pitch changes. In a second exploratory study, Anderson, Himonides, Wise, Welch, and Stewart (2012) trained a small group of adult amusics using an intervention program with a professional singing instructor over seven weeks.

Perception (MBEA scale test, computerized pitch matching) and production (perform Happy

Birthday and a self-chosen song, imitate or match a single note or melodic fragment) measures were recorded before and after the intervention. The singing intervention did not significantly improve pitch perception in the amusic group although four of the five participants improved on the MBEA scale test. In contrast, singing accuracy judgments (employing the task used by Wise & Sloboda, 2008) for the amusics' performances of "Happy Birthday" improved significantly following training.

The current report is a case study of a congenitally amusic individual who engaged in vocal training over an 18-month period. The relationship between suffering from congenital amusia and being able to enjoy music as a pastime is a complicated one. Some research reveals an expected pattern of indifference toward music (Gosselin, Paquette, & Peretz, 2015). However, there is also evidence that some congenitally amusic individuals score in the normal range on questionnaires pertaining to music appreciation (McDonald & Stewart, 2008), which indicates that they are able to engage with music. Recently, a cluster analysis looking at music-related behavior revealed that 59% of amusics (and 6% of controls) tend to avoid musical activities, while 41% of amusics (and 94% of controls) will engage in them (Omigie, Mullensiefen, & Stewart, 2012). Our case study participant both reports and scores high on enjoyment of music. This implies that he is in the minority of amusics (within the 41% as outlined by Omigie et al. (2012), who are musically engaged. Not only does he engage in musical activities such as attending concerts, he scores above average in his absorption in music score, rather than simply being in the normal range.

On the basis of prior evidence, we expected that vocal training might lead to an increase in pitch production accuracy, with concomitant changes in right fronto-temporal connectivity, from baseline. On the other hand, we made no clear prediction regarding pitch perception.

Despite the prolonged period of training and high interest in music, it is possible that pitch perception deficits are irreversible in the adult amusic brain. Beyond this assessment, we expected that it might also be possible that improvements would be transient in nature varying somehow with the extent of recent training. Through a combination of musical production and perception tasks, we examine the case of Tim Falconer², a musicophile who loves listening to music, and who often writes about music, including his own experience as a congenital amusic (Falconer, 2016).

Method

Ethics

The research protocols and collection of data were approved independently by the Ryerson University Research Ethics Board (REB) and the Comité mixte d'éthique de la recherche du Regroupement Neuro-imagerie Québec (CMER-RNQ).

Participant

Tim Falconer is a 54-year-old right-handed male who is a self-identified person living with congenital amusia. While he describes himself as tone deaf and a “bad singer”, he reports having loved music for as long as he can remember. He goes to concerts regularly – around twice a month – and listens to music daily on his mobile music player. He has specific preferences and dislikes with regard to music. He reports that he enjoys indie rock, alt-country, country, blues, R&B, and reggae, and that he does not enjoy heavy metal, hip hop, electronic dance music, and ‘current’ pop.

² Tim Falconer, our participant, requested to waive his right to anonymity.

To further characterize Tim's interests and experience with music, we administered a series of standardized tests in advance of our first test session. The scores on these tests were compared to norms using software developed by Crawford, Garthwaite, and Porter (2010) for comparing case study participants to norms (Crawford & Howell, 1998; Crawford & Garthwaite, 2002; Crawford, Garthwaite, & Porter, 2010). This software provides an estimated t -value (denoted as t_{est}), a significance value for deviation from the norm, an effect size, and an estimated percentile for the case study participant along with confidence intervals.

The Short Test of Music Preference (STOMP; Rentfrow & Gosling, 2003) assesses the types of music that an individual is interested in. Tim's results are displayed in Table 1. He scored highest and above 89.6% of the population on the reflective/complex subtest, which implies a preference for blues, classical, folk, and jazz music. He also scored above average on the intense/rebellious and upbeat conventional subtests (rock, country, pop music, etc.) but below average on the energetic and rhythmic scale (electronic music). These findings align relatively well with Tim's verbal report of musical preferences.

The Absorption in Music Scale (AIMS; Sandstrom & Russo, 2011) is a 34-item measure that assesses an individual's willingness and ability to be drawn into an emotional experience with music. The mean score on this measure is 113.5 points ($SD = 23.8$), and Tim scored 139 points. Although Tim's score was not significantly different from the center of the sample distribution ($t_{\text{est}} = 1.068$, $p = .287$), the estimated normal population below Tim's score was 85.7% [81.0, 89.6]. Based on this, we can surmise that Tim is slightly above average in terms of his emotional response to music. We also assessed Tim's ability to identify emotions conveyed by music. Tim rated each of 50 excerpts of film music with regard to how much each excerpt evokes one of five emotions (anger, fear, happiness, sadness, tenderness) on a scale of 1 to 5.

The film music excerpts were obtained from Eerola and Vuoskoski (2011). When compared to scores from non-amusic participants tested in Eerola and Vuoskoski's (2011) study, Tim performed well in terms of detecting fear, with 100% of his responses being consistent with the mean response provided by non-amusic controls. He also scored well on perceiving tenderness (70%), and happiness (60%). He was, however, somewhat impaired in detection of sadness (20%) and anger (10%), confusing the former with tenderness, while he often confused the latter with fearfulness.

Based on these demographic statistics and standardized questionnaire measures, we can surmise that Tim is not a typical amusic. He has distinct music preferences, is above average with respect to absorption in music, and he reports regular concert attendance and listening to music daily. In sum, Tim is a musicophile despite his amusia.

Comparison groups. A group of amusic participants as well as a control group of normal participants were recruited for comparison. Behavioural and diffusion imaging data were obtained from Tim, 12 additional amusic participants and 12 gender-, age-, education-, and music training-matched control participants. Participants who receive a global score on the MBEA that is over two standard deviations less than the mean are considered to be amusic. Participants were excluded on the basis of risk factors for MRI (e.g., irremovable metal in the body), factors which can adversely affect the interpretation of fMRI data (e.g., taking medications affecting the central nervous system), hearing loss, and developmental or learning disorders. Amusic participants included 9 women, were aged from 23 to 72 years ($M = 58.58$, $SD = 15.29$), and had a maximum of three years of musical training ($M = 0.92$ year, $SD = 1.11$). Control participants included 8 women, were aged from 23 to 74 years, ($M = 58.08$, $SD = 15.08$), and had a maximum of eight years of musical training ($M = 2.58$, $SD = 2.40$). There was one less

woman in the control group because one female control was matched to two amusic twins. Note that controls did have slightly more musical training than the amusia group, $t(23) = 2.09$, $p = .048$. Participants were reimbursed \$120 for the two 2-hour experimental sessions.

Timeline

Testing occurred over seven sessions, including one test session that preceded training (i.e., baseline) and another that followed training. Two of the test sessions were conducted at the International Laboratory for Brain, Music, and Sound Research (BRAMS) in Montreal, and five were conducted at Ryerson University in Toronto. Tim received vocal training from Micah Barnes, a celebrated jazz vocalist and elite vocal coach in Toronto. Their training sessions were broken down into approximately 30% vocal exercises and 20% working on specific song selections. The remaining time was split equally between vocal pitch matching, breathing exercises, talking about performance issues, talking about music in general, and talking about other topics. In addition, Tim attempted to adhere to practice between lessons, though adherence was variable. While this lack of control over practice is not ideal from a research standpoint, it was unavoidable given the reality of an individual with a very busy professional and personal schedule. However, training was continuing throughout the 18 months of the experimental timeline. Furthermore, studies in training children classified as ‘monotone’ singers found that they showed significant improvements in pitch singing after 5 sessions in 2 weeks (Joyner, 1969) or over 16 sessions in 8 weeks (Roberts & Davies, 1975), so we could not necessarily require 18 months of strict training to see results. Figure 1 shows a timeline of test sessions and the average amount of practice that Tim reported in between sessions.

Perception tests

The MBEA evaluates an individual's ability along 6 subtests – namely, scale discrimination, melodic contour discrimination, interval discrimination, rhythmic contour discrimination, metric discrimination, and an incidental memory test. Each of these subtests has been normed against 160 normal (non-amusic) participants (Peretz, Champod, & Hyde, 2003).

The Beat Alignment Test (BAT; Iversen & Patel, 2008) is a psychophysical test designed specifically to study rhythmic abilities. It includes tasks that involve synchronized tapping to beats at different tempi and meters, as well as a perceptual task. We employed the perceptual task only, in which participants are played musical excerpts along with a metronomic 'beep' at a steady beat. This beat could coincide with the beat of the musical excerpt, could contain a tempo error (too fast or too slow for the excerpt), or could include a phase error (at the correct tempo, but misaligned with the excerpt). Participants are asked to listen to the overlaid excerpt and to respond as to whether the beat is correct or incorrect.

Production and Performance tasks

The production and performance tests conducted at Ryerson and BRAMS were similar, though not identical. The specific details of each are detailed below. All analyses of pitch were conducted using Praat (Boersma, 2001). Once pitch frequencies were ascertained, they were converted into cents and then compared to the presented tone (or interval) for accuracy. In analyzing singing of *Happy Birthday*, each individual interval was calculated and compared to the 'correct' interval for each part of the song.

Ryerson sessions. Stimuli for production and performance tasks at Ryerson were presented over two KRK Rockit 5 loudspeakers using ProTools 8 software and a Digidesign 003 stimulus presentation system. Tim's singing was recorded using a Rode NTK microphone in a

double-walled recording studio with a cork floor and sound-dampening panels on the walls. In the pitch production tasks, tones were sine waves played 9.7 dB above a background pink noise floor, which was presented at 73.3 dB.

We employed an interval-matching task based on Loui, Guenther, Mathys, and Schlaug (2008). This task involves reproduction of 11 musical intervals realized using pure tones. In order to make the task more manageable, we transposed the intervals into Tim's vocal range (from the original 500 Hz starting note to a 250 Hz starting note), as transposition has been shown to be challenging for poor-pitch singers (Pfordresher & Brown, 2007). As such, the first note always had a frequency of 250 Hz, and the second note ranged between 225 and 275 Hz, in 2.5 Hz steps. In retrospect, an additional methodological consideration that could have further benefited his performance would have been to realize the intervals using a vocal timbre. Recent research has shown that reproductions with vocal timbre tends to be more accurate than non-vocal timbres (Mantell & Pfordresher, 2013; Hutchins & Peretz, 2012).

We also asked Tim to sing "Happy Birthday" from memory and without a starting pitch (as per Pfordresher & Brown, 2007; Hutchins, Larrouy-Maestri, & Peretz, 2014). We determined deviance for each interval and then averaged the absolute values of those deviances for each performance of the song. In addition to this pitch-interval analysis, we also had 14 independent raters subjectively rate the quality of each performance on a scale of from 1 to 10 (with 10 being the highest score possible). Raters were all active musicians, with an average of 17.86 years of performance experience ($SD = 5.45$) and an average of 8.78 years of formal training ($SD = 3.53$). The raters were blind as to which performance and which session was being rated at each time.

BRAMS sessions. All stimuli were presented to participants through DT 990 Pro headphones (Beyerdynamic, Heilbronn, Germany) using Max/MSP (Cycling'74, San Francisco,

CA), and the pitch productions were recorded with a TLM 103 microphone (Georg Neumann GmbH, Berlin, Germany).

In the pitch perception task, participants heard pure tone targets at five pitch heights (B3 (246.94 Hz), C#4(277.18 Hz), D#4 (311.13 Hz), F4 (349.23 Hz) and G4 (392.00 Hz) for women, and the same tones an octave lower for men). Target tones were presented randomly over 100 trials. For each trial, participants were required to move a slider that produces complex tones imitating the timbre of the human voice (see Hutchins & Peretz, 2012) to match the pitch of a pure tone target. Due to a lack of time, not all participants finished the whole task. Of the 100 trials, Tim Falconer completed 40 trials, amusics completed an average of 77.4 trials, and controls completed an average of 88.0 trials. All participants completed a minimum of 40 trials.

In the pitch production task, participants were first recorded singing the syllable /ba/ at five different self-selected pitch heights within a comfortable range for use as target tones. These targets were amplitude normalized and trimmed to remove leading and trailing silences. Target tones were presented randomly over 100 trials. For each trial, participants were required to match the pitch of the self-produced target with their voices. Almost every participant was able to complete the whole task, with at least of 87 trials for amusics and 79 for controls. Of the 100 trials, Tim Falconer completed 100 trials, amusics completed an average of 96.8 trials, and controls completed an average of 96.2 trials. Tim was also asked to sing “Happy Birthday”, once with the usual lyrics, and once with “La” sung on each syllable.

Diffusion data acquisition. All magnetic resonance acquisitions were performed on a Siemens 3T Magnetom TrioTim scanner with a 32-channel head coil at the Unité de Neuroimagerie Fonctionnelle, Centre de recherche de l'Institut universitaire de gériatrie de Montréal. T1-weighted images of the whole brain were acquired using an MPRAGE sequence

($T_R = 2300$ ms; $T_E = 2.91$ ms; FA: 9° ; FOV = 256×256 mm²; 256×256 matrix; 176 axial slices of 1 mm; acquisition time: 9 min 50 s). Diffusion-weighted images of the whole brain were acquired using a single-shot, spin-echo, echo-planar sequence ($T_R = 9.3$ s; $T_E = 93$ ms; FA = 90° ; FOV = 256×256 mm²; 61 axial slices of 2 mm; $b = 1000$ s/mm²; 64 directions; acquisition time = 10 min).

Diffusion data processing. All diffusion-weighted images were pre-processed and analysed using FSL (Jenkinson et al, 2012). Pre-processing steps included eddy-current correction (FDT Toolbox, Behrens et al., 2003) and skull-stripping (BET; Jenkinson et al., HBM 2002). DTIFIT (FDT Toolbox; Behrens et al., 2003) was used to create fractional anisotropy (FA) maps for each participant. Next, we used the methods described in detail by Chen et al. (2015) to perform probabilistic tractography of the right and left arcuate fasciculus (AF) by tracking to a target in the pars opercularis for each participant. This process involved the creation of tractography masks for each participant in MarsBar (Brett et al., HBM 2002), and data processing using BEDPOSTX (2 fibres per voxel; weight = 1, burn in = 1000) and PROBTRACKX (number of samples = 5000; curvature threshold = 0.2) (FDT Toolbox; Behrens et al., 2007). In addition to following the steps reported by Chen et al. (2015) we also applied a CSF exclusion mask to ensure that the produced tracts stayed within brain tissue. This mask was created by partitioning the participant's T1-weighted image into CSF, grey matter, and white matter using FAST segmentation (Zhang et al., 2001). The left and right AF were traceable for all participants with the exception of one amusic individual. For the remaining participants, average FA and tract volume (in voxels) were extracted for the left and right AF.

Results

Perception Tests

Tim was tested on the MBEA three times. His results are reported in Table 2. Previous research with the MBEA has shown a high level of test-retest reliability, with very few situations where individuals diagnosed as amusic are later found to be out of the amusic range. As such, any changes in scores on MBEA subscales can be attributed to improvements in ability rather than due to practice effects or change. Before training (Session 1), he scored significantly below average on scale, contour, interval, and meter. This testing session (28th March, 2011) is the first time Tim was diagnosed with amusia. In his second attempt, following the onset of training (Session 2), he scored significantly below average on contour and meter. In the post-training assessment (Session 7), he scored significantly below average on scale, contour and interval. Between sessions 1 and 2, Tim's scores on scale and interval changed from being significantly below average, to being in the normal range. However, both scale and interval returned to being significantly below average in the final assessment (Session 7), which suggests that the previous improvement was transient. We did see an improvement on the meter subscale between sessions 2 and 7, and while we cannot definitively state whether this would be a lasting improvement, the results from the BAT lend credence to this possibility.

Tim was tested on the BAT twice (Sessions 2 and 7). Estimates of significance were determined as for the demographic information. In the first assessment (Session 2), following 1-year of vocal lessons he scored significantly below the norm for correct trials ($t_{\text{est}} = -2.302, p = .029$), marginally below the norm for tempo change trials ($t_{\text{est}} = -1.762, p = .089$), and within the normal range for the phase change trials ($t_{\text{est}} = -0.243, p = .809$). Following his full training regimen (Session 7), Tim was not significantly different from the norm on any of the subscales

(correct: $t_{\text{est}} = -0.659$, $p = .515$; tempo change: $t_{\text{est}} = -0.268$, $p = .790$; phase change: $t_{\text{est}} = 0.504$, $p = .616$).

Production / Performance Tasks

Ryerson tasks. In the interval-matching task, we saw a fluctuating pattern of performance (see Figure 2). While we acknowledge that error terms are not independent (as they are all based on one singer), this analysis provided insight into relative improvements of ascending and descending intervals over time. Data were subjected to a mixed model ANOVA, with data grouped by session, and each interval serving as an individual data point. The ANOVA yielded a main effect of session, $F(3, 63) = 13.192$, $p < .001$. Multiple comparisons between time points were conducted to examine differences using Bonferroni comparisons ($p < .05$), which revealed significant differences between sessions 2 and 3, 3 and 4, and 4 and 6, respectively. However, as Figure 2 indicates these changes were a combination of improvements (from session 2 to 3, 4 to 6) and regression (between sessions 3 to 4). This pattern of data existed for 10 of the 11 intervals produced, as well as in the mean data. Interestingly, there was also a significant improvement between sessions 2 and 6, indicating that improvement is occurring overall, although this pattern of data is volatile. This pattern of results aligns with Tim's training schedule (see Figure 1), with improvement corresponding with periods of high intensity, and regression with periods that were relatively dormant. For example, Tim had no lessons and engaged in minimal practice between sessions 3 and 4, and we can see a concomitant decrease in accuracy between these sessions. Alternately, he had 10 lessons over the span of 2 months, and practiced 4 hours per week between sessions 4 and 6, and showed a large improvement.

Figure 3 displays the average deviance from correct intervals contained in "Happy Birthday" being sung with words. These data showed a marked increase in performance

between sessions 1 and 2, plateauing until session 4, and then a slight increase in session 5 followed by a regression in session 6. The increase in performance accuracy in session 5 is most likely attributable to song-specific training³. These results are mirrored by the subjective evaluations of singing quality (Figure 4; only available for Ryerson test sessions), which showed an increase in quality between sessions 2 and 3, a decrease between sessions 3 and 4, and a relatively large increase between sessions 4 and 6.

We also analysed deviation separately for ascending and descending intervals. Data were subjected to a mixed model ANOVA, with each interval serving as an individual data point. Data were grouped by interval direction (ascending vs. descending; between-items) and session (within-items). While acknowledging that the data are not independent (as performance on any interval is related to performance on other intervals) and that error terms are not independent (as above), this analysis provided insight into relative improvements of ascending and descending intervals over time. The ANOVA yielded a main effect of type, $F(1,18) = 6.739, p = .018$, as well as a main effect of session, $F(5, 90) = 4.324, p = .001$, with no significant interaction ($p = .405$). This indicates that descending intervals were performed more accurately than ascending intervals, and that Tim's performance improved on both types of intervals over the six test sessions (see Figure 5). Pairwise comparisons indicated that Tim showed statistically significant increases in accuracy between sessions 1 and 2, and between sessions 4 and 5.

BRAMS tasks. Figure 6 shows average deviance for performances of "Happy Birthday" recorded at BRAMS (sessions 1 and 5), comparing performances sung with words vs. those sung on the syllable "La". Whereas both words and "La" show a decrease in deviance, the

³ Tim reports spending a considerable amount of his practice time working on his performance of *Happy Birthday* in this period.

improvement on “La” was more substantial. In addition, while all performances with words had perfect melodic contour, melodic contour on “La” improved from 60% in the first session to 100% on the second.

Performance on each trial of the pitch perception and production tasks were coded for accuracy based on the size of the error between the target tone and slider tone or vocal tone, respectively. Trials were coded as correct when the difference between the slider tone (perception) or vocal tone (production) and the target tone was less than 50 cents, and incorrect if that difference was larger than 50 cents. Full results for Tim and both control groups are displayed in Figure 7. Before training (Session 1), Tim performed significantly better than the amusic group on the pitch perception task, $t(9) = 3.19, p = .01$, and was equivalent to the control group, $t(11) = 1.51, p < .16$. On the pitch production task, Tim performed significantly worse than both the amusic group, $t(9) = 3.43, p = .007$, and the control group, $t(11) = 21.69, p < .001$. While it was unexpected that Tim would perform at a level equivalent to normal controls (and better than amusic controls) in a pitch perception task, this may be due to a speed-accuracy trade-off. Participants were able to control the amount of time they had to do each trial, and Tim used an average of 44.56 seconds per trial. One sample t-tests of amusic and normal controls against a test value of Tim’s time reveal that he took significantly longer than both amusic ($M = 23.12$ s, $SD = 13.54$ s, $t(11) = 5.485, p < .001$) and normal controls ($M = 18.39$ s, $SD = 8.54$ s, $t(11) = 10.616, p < .001$). This also contributed to the fact that Tim only completed 40 out of 100 trials in the task overall, compared with 77.4 for amusic controls and 88 for normal controls.

Diffusion Analyses

Fractional anisotropy. Tim’s right arcuate fasciculus (AF) had significantly lower mean FA than amusics and controls, $t(11) = 3.39$ and $3.10, p = .006$ and $.01$ (see Figure 8). His left AF

had a mean FA equivalent to amusics, $t(11) = 1.08$, $p = .31$, and marginally lower than controls, $t(11) = 2.10$, $p = .06$.

Volume. Tim had a right AF (Figure 9) that was equivalent in volume to the amusic group, $t(11) = 1.13$, $p = .28$, and marginally smaller in volume than controls, $t(11) = 1.90$, $p = .09$. He had a left AF comparable in volume to both the amusics, $t(11) = 1.04$, $p = .32$, and controls $t(11) = 1.33$, $p = .20$. Figure 10 displays probabilistic tractography for the right arcuate fasciculus for Tim, as compared with a representative control brain.

Discussion

Overall, our findings indicate that 18 months of vocal training in an amusic adult does not appear to be enough to overcome long-standing deficits in pitch perception. Our production tasks showed marginal increases in accuracy of interval matching. These improvements seemed to be transient in nature, coinciding with periods where Tim was intensely focused on practice. In assessing Tim's performances of Happy Birthday, we see an increase in production accuracy through training, especially at times when he was practicing intensely. Beyond interval production accuracy, we observed improvements when blind assessors evaluated his performances. This improvement may have relied in part on aspects of the musical performance other than pitch, including rhythm.

In terms of music discrimination results, we observed an increase on the meter test of the MBEA only, after training. Before training Tim scored below average on the meter subtest, and after training he was no longer below average on meter. His performance remained within the normal range on the rhythm subtest throughout the duration of testing. These preserved rhythmic abilities are further bolstered by results from the BAT, in which Tim was significantly below the average on identifying correct tempi, and marginally below average on identifying

tempo variations before training, while he was in the normal range for identifying phase errors. Thus, we do see improvement on his rhythmic perception, as his performance on all three subtests was within the normal range after training. So while Tim was hoping to show improvement on both melodic and rhythmic perception and production, it seems that improvements were largely rhythmic in nature. However, we must also note that when Tim's performance of Happy Birthday was scaffolded by lyrics, he adhered to the musical contour of the song perfectly. When singing to "la" we saw significant improvement after training in terms of contour, and in this way we do see some degree of pitch production improvement.

This persisting difficulty with pitch-based aspects of music processing (apart from some improvement in contour) was associated to a reduction of the main white matter tract that connects the right auditory cortex to the right IFG. Fractional anisotropy in Tim's right arcuate fasciculus was significantly lower than in normal controls as well as in other amusics. Additionally, the volume of his right arcuate fasciculus tended to be smaller than that of normal controls. This finding is also highly consistent with the literature (Loui et al, 2009; Halwani et al., 2011).

Regarding the limited impact that training had on Tim's performance, it is important to consider the times that he focused on training. While he showed some improvement in singing ability when he was practicing intensely, this training did not sustain itself. With the lack of overall, long-lasting improvement, we must consider that training of adults with congenital amusia may simply not be possible. Alternatively, given that we saw hints of improvement, maybe 18 months of intermittent (i.e., non-intensive) training is not enough. It is also possible that Tim's training did not lead to sustained improvement because he has a fully matured brain. Trainor (2005) reviews the literature around the possibility of critical periods in musical

development, and finds that tonality knowledge develops in the first years of life, with harmonic knowledge needing to be acquired by around the age of 12. Steele, Bailey, Zatorre, and Penhune (2013) discuss plasticity of white matter tracts during development, noting that the arcuate fasciculus is developing during childhood, and its size and function may be influenced by musical experiences before the age of seven. Given that Tim is decades beyond this threshold, it may simply be too late for him to be successfully trained in music perception or production. Future research should build on this case study and engage in longitudinal study to test the plausibility of a critical periods argument in terms of melodic processing. If this hypothesis were to hold true, it would underscore the importance of musical education in early childhood in order to lay an important foundation for future musical study.

Another issue to consider further with Tim would be potential effects of consciousness on his amusia. Consciously attending to pitch (as Tim would have during singing training and practice) has, paradoxically, been shown to interfere with the perception of pitch in individuals with amusia (Zendel, Lagrois, Robitaille, & Peretz, 2015). When presented with errant pitches while they were monitoring a non-pitch related click, amusics and normal controls showed an early right anterior negativity (ERAN). When these pitches were presented during a pitch-monitoring task, however, amusics no longer demonstrated the ERAN response, indicating that they were not sufficiently perceiving the tones. An additional caveat related to Tim's awareness of his amusic condition pertains to potential demand characteristics in his training. Given that he was in the process of writing a book about congenital amusia, this may have affected his progress in some manner, whether conscious or unconscious. However, based on our interactions with Tim, we believe that his motivation was genuine – he came to this project initially because

of his interest in whether he would be able to learn to sing, and we have no reason to believe that he was in any way dishonest about his motivation.

Whether due to a neurological deficit, a lack of early exposure, irregular practice, or some combination of the above, what is clear is that Tim was not able to reliably improve his melodic perception or production. We did observe some improvements in his singing ability (the task he was trained on), but these improvements were transient. It is possible, as has been discussed above, that the training regimen was either too short, or lacking in the intensity required to reveal a lasting improvement. Converging evidence from perception and production tasks completed before, during and after training, at two separate institutions allows us to build a strong case against the possibility of lasting improvements in Tim's singing ability. We see some improvements in rhythmic perception, and the quality of his performances as assessed by listeners, but not sustained improvements in the pitch content of his singing (with the exception of melodic contour). As such, Tim remains a remarkable case – an amusic musicophile.

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Table 1 – Scores on Short Test of Music Preferences (STOMP)

Measure	Score	Norm	t_{test}	p	Percentile	95% CI
Reflective/complex	5.75	3.87	1.26	.210	89.6	89.0 , 90.3
Intense/rebellious	6.00	5.00	0.69	.493	75.3	74.3 , 76.3
Upbeat/conventional	4.25	3.74	0.40	.690	65.5	64.4 , 65.5
Energetic/rhythmic	4.67	3.99	-0.45	.654	32.7	31.7 , 33.8

Table 2 – Scores on Montreal Battery of Evaluation of Amusia (MBEA)

Measure	Norm	Session 1			Session 2			Session 7		
		Score	<i>t</i> _{test}	<i>p</i>	Score	<i>t</i> _{test}	<i>p</i>	Score	<i>t</i> _{test}	<i>p</i>
Scale	27	22	-2.17	.016	23	-1.73	.085	18	-3.91	<.001
Contour	27	19	-3.47	<.001	22	-2.27	.023	18	-4.10	<.001
Interval	26	18	-3.32	<.001	23	-1.25	.215	21	-2.08	.039
Rhythm	27	28	0.48	.636	28	0.48	.636	23	-1.90	.059
Meter	26	16	-3.44	<.001	18	-2.75	.007	22	-1.40	.171
Memory	27	23	-1.73	.085	26	-0.43	.665	26	-0.43	.665

Figure Captions

Figure 1. Timeline of test sessions and vocal training undertaken by Tim.

Figure 2. Average deviation (in cents) in interval matching task. Light grey lines represent individual intervals matched, black line represents mean.

Figure 3. Average deviation (in cents) in performing *Happy Birthday*.

Figure 4. Average subjective rating of quality of performance of *Happy Birthday*. Error bars represent standard error.

Figure 5. Comparison of deviation for ascending and descending intervals in performing *Happy Birthday*. Error bars represent standard error.

Figure 6. Average deviation in performance (in cents) in performing *Happy Birthday* using lyrics and singing to the syllable “La”.

Figure 7. Average percent correct on production with the slider (left panel) and production with the voice (right panel) tasks for Tim, a normal control group, and a group of amusic participants in Session 1. Error bars represent 95% confidence intervals around the mean.

Figure 8. Fractional anisotropy of arcuate fasciculus for Tim, amusic controls, and normal controls. Error bars represent 95% confidence intervals around the mean.

Figure 9. Estimated tract volume of arcuate fasciculus for Tim, amusic controls, and normal controls. Error bars represent 95% confidence intervals around the mean.

Figure 10. Right arcuate fasciculus of Tim Falconer (red) and a representative control (blue) displayed on a standard MNI brain image in coronal, sagittal, and axial planes (left to right). Tract images were generated via probabilistic tractography and thresholded at an intensity value of 50 (number of samples passing from the seed mask to the voxel).

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Disclosure Agreement

The authors acknowledge no competing financial interests.

Figure 1

Figure 2

Figure 3

Figure 4

Figure 5

Figure 6

Figure 7

Figure 8

Figure 9

Figure 10