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The Prevalence of Congenital Amusia

par
Mélanie A. Provost

Département de Psychologie
Faculté des Arts et des Sciences

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présenté par

Mélanie A. Provost

a été évalué par un jury composé des personnes suivantes :

Denis Cousineau, président-rapporteur

Isabelle Peretz, directrice de recherche

Sarah Lippé, membre du jury

L'amusie congénitale est un trouble neurogénétique qui se caractérise par une inhabileté à acquérir des habiletés musicales de base, telles que la perception musicale et la reconnaissance musicale normales, malgré une audition, un développement du langage et une intelligence normaux (Ayotte, Peretz & Hyde, 2002). Récemment, une éude d'aggrégation familiale a démontré que 39% des membres de familles d'individus amusiques démontrent le trouble, comparativement à 3% des membres de familles d'individus normaux (Peretz *et al.*, 2007). Cette conclusion est intéressante puisqu'elle démontre une prévalence de l'amusie congénitale dans la population normale. Kalmus et Fry (1980) ont évalué cette prévalence à 4%, en utilisant le Distorted Tunes Test (DTT). Par contre, ce test présente certaines lacunes méthodologiques et statistiques, telles un effet plafond important, ainsi que l'usage de mélodies folkloriques, désavantageant les amusiques puisque ceux-ci ne peuvent pas assimiler ces mélodies correctement. L'étude présente visait à réévaluer la prévalence de l'amusie congénitale en utilisant un test en ligne récemment validé par Peretz et ses collègues (2008). Mille cent participants, d'un échantillon homogène, ont complété le test en ligne. Les résultats démontrent une prévalence globale de 11.6%, ainsi que quatre profiles de performance distincts: pitch deafness (1.5%), pitch memory amusia (3.2%), pitch perception amusia (3.3%), et beat deafness (3.3%). La variabilité des résultats obtenus avec le test en ligne démontre l'existence de quatre types d'amusies avec chacune une prévalence individuelle, indiquant une hétérogénéité dans l'expression de l'amusie congénitale qui devra être explorée ultérieurement.
Mots-clés: Amusie congénitale, étude de prévalence, trouble neurogénétique.

Congenital amusia is a heritable disorder in which subjects fail to acquire basic musical abilities, such as normal music perception and music-recognition abilities, despite normal hearing, normal language abilities, and normal intelligence (Ayotte, Peretz & Hyde, 2002). Recently, a family-aggregation study showed that 39% of first-degree relatives in amusic families express the disorder, compared to 3% in control families (Peretz *et al.*, 2007). This latter finding is interesting in that it illustrates a prevalence of the disorder in non-amusic families. Kalmus and Fry (1980) evaluated the prevalence of congenital amusia at 4%, using the Distorted Tunes Test (DTT). However, this test presents some methodological and statistical problems, such as a strong ceiling effect, as well as the use of folkloric tunes, which disadvantages the amusic participants since they cannot assimilate these melodies correctly. The present study aimed at re-evaluating the presence of congenital amusia, using a recently validated online test by Peretz and colleagues (2008). One thousand one hundred participants, from a homogeneous sample, completed the online test. Results showed a global prevalence of 11.6%, with four distinct patterns of performance emerging: pitch deafness (1.5%), pitch memory amusia (3.2%), pitch perception amusia (3.3%), and beat deafness (3.3%). The variability in the results obtained with the online test brings evidence of at least four types of amusias with individual prevalences, indicating a heterogeneity in congenital amusia that needs to be further explored in later studies.

Key words: Congenital amusia, prevalence study, neurogenetic disorder.

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The Prevalence of Congenital Amusia

Humans are born with the potential to both speak and make music (Mithen, 2005). It seems to emerge instinctively in all known human societies (Peretz, 2006), as well as develop spontaneously in children, without a conscious effort to do so and without formal instruction. For example, even before babies can speak, they display certain musical abilities similar to adults such as sensitivity to musical scales as well as to temporal regularity. This is observable through processing consonant rather than dissonant intervals more easily (Schellenberg & Trehub, 1996), showing a learning preference for scales with unequal steps (Trehub, Schellenberg & Kamenetsky, 1999), and by presenting a preference for music with an isochronous pulse (Drake, 1998).

Recently, it has been shown that despite the apparent universality of music, certain individuals fail to acquire these basic musical abilities, notably pitch perception, and that these difficulties might have a neurogenetic origin (Peretz, Cummings & Dubé, 2007). This musical deficit has been called note-deafness (Allen, 1878), tone deafness (Fry, 1948), tune deafness & dysmelodia (Kalmus & Fry, 1980), and most recently congenital amusia (Peretz, 2001). All of these terms refer to the same condition, whereby adults who report lifelong difficulties with music exhibit a deficit in detecting pitch changes in melodies. The term "amusia" seems preferable, to acknowledge the possibility that there exist as many forms of congenital amusias as there are forms of acquired amusias that are the consequences of accidental brain damage (Stewart, von Kriegstein, Warren &

Griffiths, 2006). The term “congenital” means only present from birth; it defines a likely time period but not the etiology.

Thus, it is likely that musical ability, like language capacity, has a genetic component that helps guide neural growth to better facilitate both language and music processing. However, it would appear that abnormal development of both language and music abilities sometimes occurs. In the speech domain, such conditions are often termed “specific language impairment” and a large research effort has been undertaken to understand the origins and varieties of these disorders (Bishop & Snowling, 2004). Specific language impairment (SLI) is clinically defined as failure to develop language normally, given adequate environment for learning language and the absence of hearing deficits, mental retardation, oral motor/structural abnormalities, and neurological or psychiatric impairments affecting language acquisition (Bartlett *et al*. 2002). Familial aggregation studies and twin studies suggest that SLI has a genetic component (for a review, see Stromswold, 1998). These studies show a significantly increased incidence of impairment in first-degree relatives in families containing a proband (18%–42%) versus control families (3%– 26%). Further, the prevalence of SLI is estimated between 3% and 10% (Tomblin *et al.*, 1997), similar to the prevalence of developmental dyslexia (Snowling, 2000), which has been linked to SLI.

In the musical domain, music-specific impairments have been reported (Peretz & Hyde, 2003). All reports of congenital amusia document a musical disorder that appears to be remarkably similar across cases. Subjects fail to acquire basic musical abilities, such as normal music perception and music-recognition abilities, despite normal hearing, normal language abilities, and normal intelligence (Ayotte, Peretz & Hyde, 2002).

Congenital amusia appears to be not only specific to the musical domain but also to be monosymptomatic (or nonsyndromic), because there is no parallel neurodevelopmental disorders such as dyslexia, autism or specific language impairment (Peretz, Cummings & Dubé, 2007). These individuals have a normal understanding of speech and prosody, they can recognize speakers by their voices and can identify all sorts of familiar environmental sounds, such as animal cries. What is specific to amusic individuals is their inability to recognize a familiar melody without the aid of the lyrics and their failure to detect out-of-tune singing, including their own (Peretz & Hyde, 2003). Most notably, they fail to detect out-of-scale notes in conventional but unfamiliar melodies (Ayotte *et al.*, 2002). What amusics seem to be lacking are the implicit knowledge and procedures required for mapping pitches onto musical scales.

Further, these seem to aggregate in families (39 %) (Peretz, Cummings & Dubé, 2007). Its prevalence has been quantified by direct auditory testing of members of large families of amusic probands, as well as members of control families. All participants were tested with three conditions. First, the “out-of-time” condition, which consisted of introducing a silence of 5/7 of the beat duration (i.e., 143 ms) directly preceding the critical tone (the first downbeat in the third bar of the four-bar melody), thereby locally disrupting the meter (i.e., regularity). This was followed by the “out-of-tune” condition, in which the change consisted of a mistuning by half a semitone, hence introducing a “sour” note, and finally the “out of key" condition, where the change consisted of a tone that was outside the key of the melody, hence introducing a "foreign" or "wrong" note. In each condition, subjects were presented with 24 melodies (12 congruous and 12 incongruous) one at a time, in a random order. Their task was simply to detect whether an incongruity occurred in each melody and to click a "yes" button whenever they detected an anomaly and a "no" button when they did not detect an incongruity. In the family aggregation study, the amusic threshold was set at 2 SD from the control mean averaged over the two pitch conditions (comprising 24 melodies with no incongruity and 24 melodies with either a mistuned or an out-of-key pitch). Results supported the idea that congenital amusia is a heritable disorder, since 39% of first-degree relatives in the amusic families expressed the disorder, compared to 3% in control families (Peretz *et al.*, 2007). This finding is interesting, in that it shows that there is a prevalence of congenital amusia occurring in control families.

The prevalence of congenital amusia was first studied using an instrument called the Distorted Tunes Test (DTT) that determined participants' ability to remember a melodic line and judge whether it was rendered correctly. To do so, wrong notes were introduced into popular melodies without changing the rhythm, and participants were asked to identify the errors. The hypothesis was that normal adults who had been exposed to these melodies would be able to compare the stimuli to their memory of the tonal patterns, enabling them to detect the errors. Amusics, on the other hand, would not perceive the errors because their ability to detect the tonal patterns would be compromised. 600 individuals took part in the study, all raised in Britain and exposed to the popular melodies used as the stimuli.

Amusic participants were identified when an individual made three or more errors, either by false positives (i.e. stating there is an error when there is no incongruity) or by not identifying an incongruity that was present. Of those participants, 4% performed as poorly as did 20 adults who considered themselves or were considered by others to be amusic, suggesting that 4% of the population may suffer from a defect in perceiving musical pitch (Kalmus & Fry, 1980).

However, the DTT presented several methodological and statistical problems. First, the use of familiar melodies can be a problem when testing amusic participants, since their deficit impedes their ability to assimilate these melodies in the first place, putting them at a disadvantage when asked to identify out-of-key notes. Also, the majority (78.5%) of participants achieved perfect results, scoring 100%, pointing to a lack of sensitivity of the test to the presence of a disorder.

In order to better evaluate the prevalence of congenital amusia, an online auditory test based on the one used in the family aggregation study (Peretz *et al*, 2007) was designed by our laboratory which aims at uncovering individuals who have difficulties detecting out-of-key pitches in a melodic context (Peretz *et al.*, 2008). This test is an improvement over the DTT in four ways. First, it uses unfamiliar, novel melodies, designed specifically for the online test, which eliminates the confound of lack of exposure to the stimuli. Second, the scores' distribution is sensitive to the extremes, illustrating its capacity to tap all levels of musical ability. Third, and unlike the DTT, it includes a control condition which consists of presenting the melodic stimuli, into which rhythmic, but not melodic, incongruities have been inserted. This control condition rests upon past research on amusics' ability to detect off-beat changes or asynchronies (Hyde & Peretz, 2004), and allows to disregard general auditory difficulties as the source of the pitch deficit. Finally, the online test was validated using the Montreal Battery of Evaluation of Amusia, or MBEA (Peretz, Champod & Hyde, 2003), which constitutes the primary tool used to identify congenitally amusic cases across many laboratories. Further, it provides an index of musical abilities that are normally distributed and that is reliable on test-retest.

Therefore, the purpose of the present study was to re-evaluate the prevalence of congenital amusia in the general population, currently estimated at 3% - 4% (Peretz *et* al., 2007, Kalmus & Fry, 1980), using the recently validated online test (Peretz *et al*., 2008). Participants retained for the study needed to be between the ages of 18 and 40 years, and they had to be currently completing or had achieved a Bachelor's degree or higher level of education. This contributes to maintaining a more homogeneous sample.

It is predicted that the MBEA Scale test as well as the online Out-of-Key tasks will be correlated with each other, since they appear to tap the same cognitive abilities. The MBEA Scale test is used in the online test because it alone has been shown to be adequate in detecting the presence of amusia, as well as being a diagnostic test used in several laboratories. Peretz *et al.* (2008) showed that participants confirmed with the full MBEA as well potential amusics screened using only the MBEA Scale task showed similar performance outcomes in the online pitch conditions. As such, it is believed that the MBEA Scale test taps into the same cognitive abilities as the online Out-of-Key task, because both involve the insertion of an out-of-key note in a conventional melody. Finally, the MBEA Scale task also correlated with the online Out-of-Key condition (Peretz *et al.*, 2008).The MBEA Scale task is more demanding on memory than the online Out-of-Key task, since the participant must keep the first melody in their memory in order to compare it to the second one to make their judgment as to whether they were the same or different (Peretz *et al*, 2008). Further, neither pitch-based test should correlate with the online Offbeat task, since this task is designed to tap into rhythmic abilities rather than pitch-based abilities. These abilities have been shown to be separate since amusic individuals perform poorly on pitch-based tasks, but perform normally on rhythmic-based ones (Hyde & Peretz, 2004).

The meaningfulness of the 4% prevalence of congenital amusia was recently discussed by Henry & McAuley (2010). They propose that prevalence estimates depend on the specific test, cut-off, and degree of skew in the distribution. As such, it is important to keep in mind that an established prevalence is a statistical value that is test-dependent, and this applies to any study that evaluates prevalence. Although Henry & McAuley state that there is no solution to this problem for test-based methods, they propose that looking at separate scores rather than composite scores as well as looking at questionnaires would allow prevalence studies to be based on theoretically-defined patterns of performance across tests for diagnosis. As such, the prevalence would be established on results of individuals who fall below cut-off scores and who show similar profiles.

This means that any cut-off score used to establish prevalence is a statistical criterion. It is thus hypothesized that at least 2.1% of individuals will be identified as amusics, and at least two types of amusia will emerge. Further, beat-deaf cases should be uncovered as well, and their profile will be described for the first time.

Methods

Participants

Over three thousand individuals visited the website. However, not all participants completed all three tests of the online test, some did not fulfill the required criteria (age, education), and were thus eliminated. Seventy-seven participants were eliminated for other reasons: 29 reported cerebral-vascular accidents or head trauma, 36 reported audiological difficulties, one participant appear to have not understood the *Out-of-Time* task (25% success), 10 participants failed the catch trial in the MBEA scale test, and one failed all three tests including the catch trial. The final sample consisted of 1 100 individuals between the ages of 18 and 40, who were pursuing or had obtained at least a Bachelor's degree, and were unselected for their musical abilities. They were comprised of 468 males and 632 females, with an average age of 24 years and an average of 17 years of education.

Participants were recruited at both the University of Montreal as well as Bishop's University through student mailing lists, classroom visits, advertisements on the university websites, and student newspapers. Participants were also recruited from the general population through local and national newspapers as well as through local radio stations. Participants were informed that a compensation of 15$ gift certificates for music downloads would be given to one of every 50 participants.

Materials

The most recent version of the online test consists of three tests. The first test is the MBEA scale test (Peretz *et al.*, 2003), which is comprised of 30 pairs of melodies, presented with a piano timbre, composed according to Western tonal-harmonic conventions, as well as a catch trial. The first melody in the pair is unaltered. Participants are then presented with the second melody. In half of the sequences, a key-violated alternate melody was created by modifying the pitch of one tone so that it was out of key, while maintaining the original melodic contour. The remaining sequences were unaltered, meaning the same melody was presented twice (see Figure 1A and B). The catch trial involved an alternate melody in which one full measure contained pitches that were randomized over several octaves. All stimuli were generated with a piano sound.

The second and third tests (Peretz *et al.*, 2008) were constructed using 12 melodies from the MBEA scale test, all in a major mode according to Western tonal-harmonic conventions. They contain 9.6 successive tones, on average, and are computer generated at a tempo of 120 beats/min and played with a piano timbre. The 12 melodies were modified so that the same critical tone was altered either in terms of time or pitch (see Figure 1C). The tone to be changed always fell on the first downbeat in the third bar of the four-bar melody (hence, was metrically stressed) and was 500 ms long. The time change (Figure 1D) consisted of introducing a silence of 5/7 of the beat duration (i.e., 357 ms) prior to the critical tone, thereby locally disrupting the meter or introducing an offbeat tone. In the Out-of-Key condition, the change consisted of using a tone that was outside the key of the melody, hence introducing a “foreign” or “wrong” pitch in the musical context (Figure 1E). The melodies were presented with 10 different timbres (e.g., piano, saxophone, clarinet, recorder, harp, strings, guitar) to make the auditory test more interesting.

After the auditory test, participants were presented with a questionnaire comprised of questions regarding their musical background and personal history. This included basic demographic questions (e.g. age, gender and education level), as well as questions about any disorders or cognitive deficits the person might have (e.g. dyslexia or memory problems), their musical habits (e.g. frequency of music listening and musical activities such as singing and dancing), their feelings about their musical abilities, their musical environment, as well as their musical background (e.g. training and practice).

The online test can be found at this web address: http://www.brams.umontreal.ca/amusia-general/. This test is currently available online. However, at the time of testing, participants were given a code in order to access the online test.

Procedure

Once they were logged on, participants could choose their language of preference (English or French), and a short introduction to the study was presented. It was also specified that the test needed to be fully completed to be eligible for compensation. Participants were then given the appropriate code to access the online test, were informed of the testing procedure, and were asked to give informed consent by clicking on a button. Finally, participants were prompted to test their audio equipment and adjust their volume by listening to three short musical excerpts and indicating if they had heard it by clicking on a button. Thus, there was no requirement or control for neither speaker quality nor loudness of the stimuli.

Participants then began the online test. Before each test, participants received two examples of the task with feedback to ensure they understood the instructions. They were first tested with the MBEA scale test, followed by the Out-of-Time and Out-of-Key conditions. In each test, participants could answer by clicking on a same/different response for the MBEA scale test, or a congruous/incongruous response for the Out-of-Time and Out-of-Key conditions. The auditory test lasted approximately 20 minutes. After the test, participants filled out the demographic questionnaire, and received their test scores. The whole procedure was completed in roughly 30 minutes.

The cut-off that is most commonly used in psychological testing is 2 SD below the mean, which establishes a 95% confidence interval around the obtained results. The MBEA uses a composite score that is -2SD from the mean as the cut-off score for determining normal participants from amusics; therefore, the same cut-off is used in the online test. However, contrary to the MBEA and in line with Henry & McAuley's suggestion, the three separate scores of the online test were considered, and individual profiles were established according to the different patterns of performance obtained.

Results

The distribution of the raw scores on the MBEA Scale test $D(1100) = 4.79$, $p < .001$, the Out-of-Time test $D(1100) = 5.46$, $p < .001$ as well as the Out-of-Key test $D(1100) = 5.62$, $p < .001$ were all significantly negatively skewed when evaluated with a Kolmogorov-Smirnov test, illustrating most participants' ease with the online test (see Figure 2). This skew is similar to results obtained previously (Peretz *et al.*, 2008, 2003). According to Henry and McAuley, this skew would lead to an overestimate of the prevalence of congenital amusia.

One hundred and twenty six participants scored below the established cutoff score (-2SD from the mean) on at least one of the tests, and were identified as the amusic group (see Table 1). A chi-square analysis revealed no significant gender differences between the amusic group and the non-amusic participants $X^2(1) = .34$, *n.s.* Further, independent-samples t-tests revealed no significant differences for age $t(1098) = .09$, *n.s.*, as well as for years of education $t(1098) = .34$, *n.s.*

There were no significant differences between the amusic group and the non-amusic group with regards to any other neurological disorder, such as dyslexia $X^2(1) = .54$, *n.s.,* attention $X^2(1) = 1.64$, *n.s.*, memory $X^2(1) = .28$, *n.s.*, elocution $X^2(1) = .21$, *n.s.*, mathematical ability $X^2(1) = .28$, *n.s.* or orientation $X^2(1) = .42$, *n.s.* (see Table 1).

Not surprisingly, an independent samples t-test revealed a significant difference for years of musical training, $t(783) = 3.37$, $p < .001$, with the non-amusic participants displaying a few more years of training ($M = 8.9$. years) than the amusic participants ($M = 6.2$ years). It is important to note that only 72 amusic and 719 non-amusic participants reported the years of musical training they received. Significant correlations between performance on the three tasks and musical education confirmed the relation between musical lessons and test outcomes.(MBEA Scale and Musical Education $r(785) = .08$, $p < .05$, Out-of-Time and Musical Education $r(785) = .12$, $p < .05$, Out-of-Key and Musical Education $r(785) = .20$, $p < .05$).

With regards to the type of musical training, there were some significant differences between the amusic group and the non-amusic group. Although both groups had equivalent levels of musical training in school $X^2(1) = 01$, *n.s.*, amusics displayed significantly less musical training than non-amusic participants in the four other types of musical education. They had less optional lessons in school $X^2(1) = 18.4$, $p < .001$, private music lessons $X^2(1) = 19.7$ $p < .001$, conservatory classes $X^2(1) = 5.7$, $p < .05$, and had less self-taught experiences $X^2(1) = 27.9$, $p < .001$. This is not surprising in that amusic participants might not be encouraged to pursue their musical education because of their deficit (Peretz *et al*, 2008).

An ANCOVA was conducted on all three test results (MBEA Scale, Out-of-Key and Off-beat) for all types of amusias combined, with years of musical experience as the covariate. The ANCOVA revealed that the scores on the MBEA Scale test $F(2, 782) = 79.1$, $p < .001$, $\eta^2 = .17$, the Out-of-Key test $F(2, 782) =$

75.8, $p < .001$, $\eta^2 = .16$, and the Out-of-Time test $F(2, 782) = 120.9$, $p < .001$, $\eta^2 = .24$, remained significantly different between amusics and non-amusics after controlling for musical training, indicating that although musical training was significantly correlated with performance, it is not the only contributing factor to the lower scores seen in the amusic group.

It was hypothesized that the majority of amusic participants would fail on both the MBEA scale test and the Out-of-key test. However, amusics who failed on both melodic tests constituted the smallest proportion of amusics (1.5%). It was more common for participants to fail either the MBEA scale test (3.2%) or the Out-of-Key test (3.3%) separately. Further, for the Out-of-Time test, 3.3% of the sample failed this task (at -2SD as well). Only 3 participants who showed a deficit on the Out-of-Time test also failed on Out-of-Key tasks but not on the MBEA scale (see Figure 3). Accordingly, a global prevalence of congenital amusia of 11.6% was established, with four distinct profiles (see Table 2).

One-way ANOVAs between the four established types of amusias revealed that there are no significant age differences $F(4, 121) = 2.01$, *n.s.*, differences for years of education $F(4,121) = 1.93$, *n.s.* or for years of musical training $F(4,121) = .31$, *n.s.* Interestingly, there is a significant effect for gender $F(4,121) = 2.73$, $p < .05$, in which there are more men than expected who fail the Out-of-Key task only $X^2(4) = 10.4$ $p < .05$ (Table 2). Even though the number of years of musical training did not differ significantly between the amusic groups, the type of musical training received did display some significant differences (see Table 5). Five types of musical training were considered: obligatory classes given in school, optional lessons in school, private lessons, conservatory classes and self-taught musical abilities. Significant differences were found between the different profiles of amusics. These differences will be mentioned in each respective type.

In the demographic questionnaire, four questions have been identified as being relevant for the identification of amusic individuals (Peretz *et al.*, 2008, see Table 3). These questions have been shown to differentiate amusic individuals from normal participants as described below.

a. Pitch deaf amusics

Sixteen participants failed both melodic tasks of the online test, the MBEA Scale task as well as the Out-of-Key task. They exhibit a profile that is similar to the previously established one for congenital amusics (Peretz *et al.*, 2008, 2003), their answers to the diagnostic questions and statements being very similar to those of the confirmed amusics. Chi-square analyses also show that Pitch memory amusics show significant differences on all four of the diagnostic questions when compared to non-amusics: They report that they cannot recognize a melody without lyrics $X^2(1) = 25.3$ $p < .001$, cannot perceive when someone sings out-of-tune, $X^2(1) = 38.8$ $p < .001$, cannot perceive when someone produced a wrong note $X^2(1) = 66.6$ $p < .001$, and report singing out of tune $X^2(1) = 18.5$ $p < .001$.

b. Pitch memory amusics

Thirty-five participants failed only the MBEA scale task, while displaying unimpaired abilities in the Out-of-Key task. They show significant differences on all diagnostic questions $X^2(1) = 26.8$ $p < .001$, $X^2(1) = 17.0$ $p < .001$, , $X^2(1) = 23.7$ $p < .001$,and $X^2(1) = 19.5$ $p < .001$. Pitch memory amusics also have significantly less obligatory school lessons than the other profiles $X^2(3) = 9.6$, $p < .05$

c. Pitch perception amusics

Thirty-six participants failed only the Out-of-Key task while succeeding on the MBEA Scale task. They also show significant differences on all the diagnostic questions when compared to non-amusics: $X^2(1) = 39.4$ $p < .001$, $X^2(1) = 69.7$ $p < .001$, $X^2(1) = 50.0$ $p < .001$, and $X^2(1) = 27.1$ $p < .001$, (see Table 3).

d. Beat-deafness

These participants fail only the Out-of-Time component of the online test. Interestingly, they display a very different profile from the three other profiles. They display normal performance in their pitch abilities , and self-report fewer difficulties in both perception and production of pitch. This may indicate a possible form of congenital arrhythmia. Beat deaf participants tend to show the same pattern of responses as normal participants do, with no significant differences on any of the diagnostic questions. There were some questions in the demographic questionnaire that could be interesting with regards to rhythmic ability, notably *Do you dance? Do you consider yourself to be a good dancer? Can you dance?* and *I cannot follow a musical rhythm.* Chi-square analyses were conducted using these questions; however, there was no clear-cut emerging pattern from the results. Beat deaf participants have significantly more private lessons $X^2(3) = 11.0$, $p < .05$ as well as self-taught musical abilities $X^2(3) = 25.3$, $p < .001$, than the other types of amusia.

Family aggregation was evaluated through self-report of parents or siblings with musical problems (see Table 6). There were only two participants, one pitch memory amusic and one beat deaf amusic, who reported both parents displaying a musical impairment. Further, sibling data was reported sporadically and as such made drawing conclusions difficult. Family aggregation was very difficult to evaluate, because a majority of participants in most cases reported not knowing if their parents had musical difficulties. Interestingly, pitch-deaf amusics were the ones who reported most often not knowing if one or both parents had a difficulty. This trend could in fact be due to their severely impaired pitch abilities, making it impossible to evaluate the musical abilities of others.

An interesting question to be examined was the differences in musical habits between amusic participants and non-amusics. Three questions on the demographic questionnaire addressed these habits: *Do you intentionally listen to music? Do you sing in private?* and *Do you sing in public?* Chi-square analyses were conducted, and revealed significant differences between amusics and non-amusics on these three questions $X^2(1) = 6.88$,
$p < .01$, $X^2(1) = 14.02$, $p < .01$, and $X^2(1) = 4.97$, $p < .05$, respectively, where amusics display these behaviours significantly less frequently than non-amusics (see Table 7).

When the amusic types are compared with each other, the picture changes somewhat (Table 8). *Do you intentionally listen to music?* and *Do you sing in private?* remain significant $X^2(4) = 10.1$, $p < .05$ and $X^2(4) = 18.23$, $p = .001$, respectively. Interestingly, beat deaf and non-amusic participants show a similar pattern of responding to the question *Do you sing in private?*, where there are more individuals reporting that they sing in private frequently. Conversely, pitch-based amusic participants show the opposite pattern, where more participants report singing rarely. However, the responses to the question *Do you sing in public?* are no longer significantly different $X^2(1) = 7.21$, *n.s.* It can be observed that a large majority of participants do not sing in public on a regular basis.

Discussion

The purpose of the present study was to reevaluate the prevalence of congenital amusia in the general population using the recently validated online test. Results revealed a global prevalence rate of 11.6%, which can be subdivided into four distinct patterns of performance: three pitch-related amusias, as well as one form of beat deafness. The three pitch-based amusias were referred to as pitch memory amusia, pitch perception amusia, and pitch-deafness.

The first profile of pitch-based amusia is founded on deficits in pitch memory, and affected 3.2% of the sample. Gosselin, Jolicoeur and Peretz (2009) recently discussed the role of memory in congenital amusia. In their study, participants were presented with two tasks: the first was to compare two single tones separated by a retention interval and to decide whether they were the same or different, with an inter-tone (retention) interval that was either empty (no interfering tones) or it was filled with 6 distractor tones (interference). The second task was a pitch sequence task, in which two sequences of tones varying in pitch were presented, separated by a 2 second silent retention interval, and in which participants were asked to decide whether the two sequences were the same or different. The length of the sequences varied between 1, 3, or 5 tones. Results showed that amusics' performance on both tasks was impaired when compared to controls, most notably in the simple conditions where only one tone was presented. The authors concluded that for these amusics, their pitch memory difficulties are characterized not only by a difficulty in retaining pitch over time, but also by a greater susceptibility to interference from memory load. The pattern of results obtained by this group of amusic participants is supported by this evidence: they only failed the MBEA Scale task, which requires participants to keep in memory a first melody in order to compare it to a second one, allowing them to decide if both sequences are the same or different.

The second type of pitch-based amusia stemmed from pitch perception difficulties, and affected 3.3% of the sample. Foxton, Dean, Gee, Peretz & Griffiths (2004) explored this underlying deficit by assessing fine-grained pitch perception as well as the perception of more complex pitch patterns. They did this by exploring the performance of amusic individuals using tasks that tapped into three different levels of neural processing. First, participants' ability to detect pitch differences between two tones was evaluated, by assessing their ability to differentiate between two different pitches, as well as changes in pitch direction.

Second, participants' ability to detect changes in pitch sequences was assessed by asking participants to compare pairs of four-note pitch sequences, deciding whether the pairs were the same or different. The altered sequences were either contour-violated, showed pitch changes while respecting contour, or transposed by half an octave and contour-violated. Finally, the organization of sounds into perceptual streams on the basis of pitch was evaluated. Previous research has shown that small pitch separations between high and low tones give rise to a unified percept of one perceptual stream, such that it is possible to follow the triplet rhythm (Bregman, 1990). Participants were asked to identify when they could no longer hear the triplet rhythm, because this relies heavily on pitch perception since larger pitch separations eliminate the unified percept, forming two separate streams. Results showed that amusic participants showed marked difficulties in low-level pitch processing abilities, such as the ability to detect pitch differences as well as differences in pitch sequences. As such, the authors concluded that the results demonstrated pitch perception deficits in congenital amusia, both at the level of detecting fine-grained differences in pitch, and at the level of perceiving patterns in pitch. This evidence supports results obtained in the current study, since these participants only failed the Out-of-Key task, in which the incongruent condition consisted of using a tone that was outside the key of the melody, hence introducing a "foreign" or "wrong" pitch in the musical context. As such, participants' ability to detect a difference in the pitch sequence was compromised by their difficulties in pitch perception. However, their ability to succeed on the MBEA scale test could be linked to their use of other pitch cues that they are more sensitive to, such as absolute pitch or pitch range.

Finally, a third type of pitch-based amusia was identified as pitch deafness, affecting 1.5% of the sample. In this condition, participants failed both the MBEA Scale task as well as the Out-of-Key task. This form of amusia represents the rarest cases. Peretz *et al.* (2003) concluded that the pattern of performance obtained on the MBEA indicated a core deficit that is related to a basic pitch perception deficit. Amusics that have been studied using the MBEA (Ayotte *et al.*, 2002, Peretz *et al*., 2003, 2007, 2008, Gosselin *et al.*, 2009) usually show MBEA composite scores that lie below cutoff scores (-2SD), showing marked pitch impairment. Because these participants fail multiple pitch-related tasks, it would be fair to assume that these participants' pitch perception is so severely impaired that it prevents them from assimilating the melodies in order to keep them in memory, as originally proposed by Kalmus & Fry (1980). This could explain participants' poor performance on both melodic tasks in the online test.

Interestingly, a fourth distinct pattern of performance emerged with regards to participants' performance on the rhythmic portion of the online test, with 3.3% of the sample failing only this task. Past research has shown that rhythmic ability, most notably our ability to synchronize our body movements to rhythm, might be as ingrained as our language and pitch abilities (Phillips-Silver and Trainor (2005).

However, there is also some evidence that rhythm impairment may not be isolated, but could be linked to pitch impairment as well. Dalla Bella & Peretz (2003) have shown that certain confirmed congenitally amusic individuals also show impairment in their ability to tap in time to music. In their study, amusic participants and their matched controls were asked to tap in time to three musical excerpts. Results showed significant impairment in the amusics' average performance when compared to their matched controls on musical excerpts, but not when they were asked to tap in time to isochronous sequences of noise bursts. This could explain the results obtained by three amusic participants in the online test who failed the Out-of-Key and rhythmic tasks simultaneously.

Because there was only one measure of rhythmic skill in the online test, it would be interesting to have a more thorough evaluation of these participants' rhythmic abilities using recently validated tests such as the Beat Alignment Test (Iversen & Patel, 2008). This tool measures a participant's ability to synchronize tapping to a musical beat, as well as their ability to perceive the beat correctly. Beat synchronization is evaluated by looking at a participant's performance on the inter-tap (finger tapping) interval (ITI) compared to the musical beat's inter-onset interval (IOI). The closer the ITI is to the IOI, the better the participant's performance, because it illustrates their ability to faithfully maintain the beat. Beat perception, on the other hand, is evaluated by presenting a musical track on which a click track is superimposed, and participants are asked to judge whether the click track is on the beat or not. The click track conditions are either truly on the beat, or are offbeat by being either off tempo or of wrong phase. This tool is interesting in that it assesses different components of rhythm ability, in the same way that the online test appears to tap into different components of musical ability.

The variability in the results obtained with the online test, especially the evidence of at least four types of amusias with individual prevalences, indicates a heterogeneity in congenital amusia that needs to be further explored. This heterogeneity in the expression of a disorder can be observed in many other neurogenetic and neurodevelopmental disorders such as Specific Language Impairment, Dyslexia, and Autistic Spectrum Disorders. By going back to the model of music perception and memory proposed by Peretz *et al.* (2003), it is possible to explore which different cognitive processes could be contributing to the different forms of congenital amusia (see Figure 4). Pitch memory amusics could be experiencing difficulties in the "Repertoire" component, where they may be having a difficulty learning the new melodies, therefore preventing them from comparing the two melodies in the MBEA Scale task. Pitch perception amusics might be experiencing impairment in only a subset of the "Melodic Organization" component of the model, such as the contour component, which is responsible for one's ability to extract pitch direction. This could explain their seeming inability to process fine-grained pitch changes. Pitch-deaf amusics might have severely impaired "Melodic Organization", which normally allows the representation of melodic contour and processes tonal information. This could interfere with their ability to properly represent interval information, preventing the emergence of tonal knowledge such as scale structures, musical keys and tonal functions. Finally, the model also proposes a separate processing system responsible for temporal organization, which could explain Beat-deafness. The model proposes a double dissociation between rhythm (the ability to group events according to temporal proximity) and meter (the extraction of an underlying temporal regularity or beat), the latter being more accurately measured by the Out-of-Time task of the online test. Beat-deafness remains the form of amusia that has been the least studied in the literature, but that seems to affect an important percentage of the population.

As such, it is important to further study these four different types of congenital amusia separately and extensively, in order to better understand the complexity of the disorder. This would also allow for a more accurate estimation of each type's prevalence, as well as a better understanding of the characteristics of each type.

Figures

Figure 1. Examples of stimuli from the online test. A) Original melody from the MBEA Scale task. B) Alternate melody from the MBEA Scale task. C) Congruent melody from the online test. D) Melody with an incongruent rhythm from the *Out-of-Time* task. E) Melody with an incongruent pitch from the *Out-of-Key* task.

Figure 2. Distribution of the raw scores on the A) MBEA Scale task, as well as the online B) Out-of-Time and C) Out-of-Key tasks.

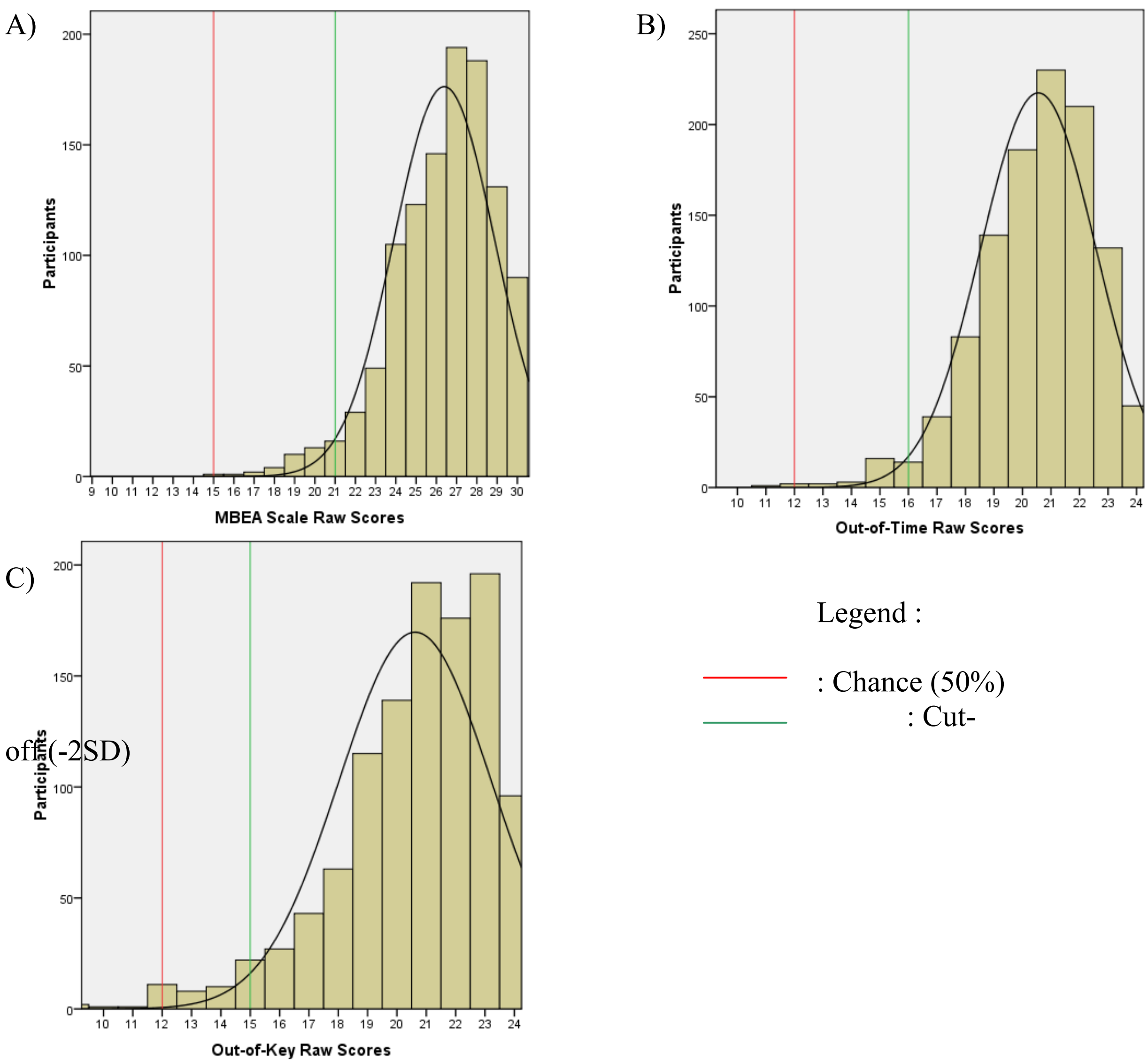

Figure 3: Distribution of Amusics by Type

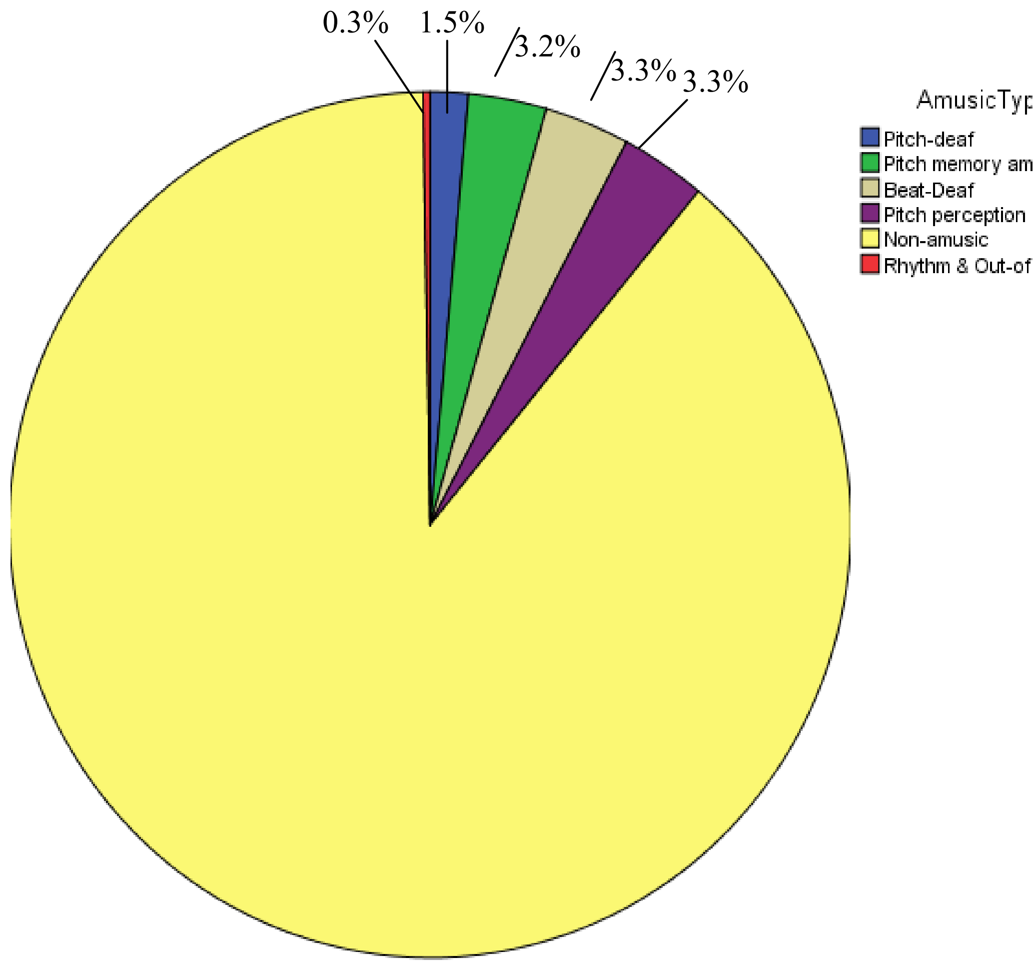

Figure 4: Model of Music Perception and Memory

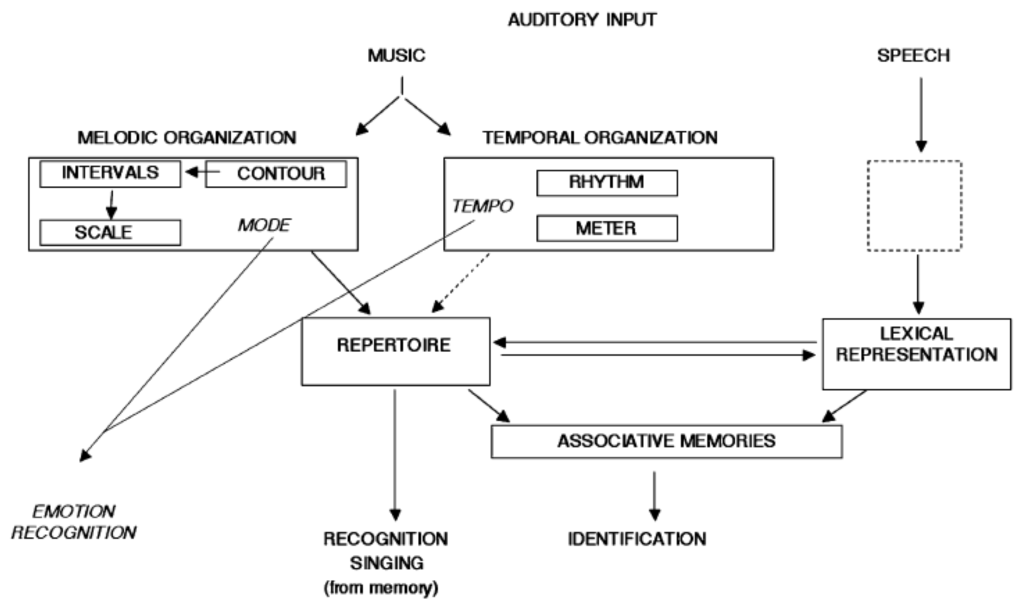

Table 1: Demographic Characteristics of the Amusic and Normal Participants

	Amusics (n = 126)	Non-Amusics (n = 972)
Gender	76F / 50M	554 F / 418 M
Age (Range)	24.2 (18-40	24.1 (18-40)
Years of Education (*Range)	16.9 (2-6)	16.8 (2-6)
Years of Musical Education (Range)	6.2 (0-22)	8.9 (0-31)
English-speakers	58 (46.0%)	586 (60.3%)
French-speakers	68 (54.0%)	386 (39.7%)
Dyslexia	4.0%	5.1%
Attention Problems	10.3%	13.9%
Memory Problems	9.5%	10.5%
Elocution Difficulties	8.0%	6.4%
Problems in Math	11.1%	11.9%
Spatial Orientation Problems	6.3%	4.6%

* 2 = Some undergraduate completed
(students outside of Québec who have completed high school – grade 12)
3 = Some undergraduate completed
(Québec students who have completed Cégep education)
4 = Bachelors completed
5 = Masters completed
6 = Doctorate completed

Table 2: Demographic Characteristics and Prevalence of the Four Profiles of Amusia, the Mean Score (SD) on each test

	Gender	Age (Range)	Education (*Range)	MBEA Scale	Out-of-Time	Out-of-Key	Prevalence
Pitch memory	22 F / 13 M	23.9 (18-40)	16.5 (2-6)	65.6% (5.3)	83.6% (7.0)	80.6% (8.5)	3.2%
Pitch perception	15 F / 21 M	24.0 (18-40)	17.3 (2-6)	83.2% (7.1)	84.0% (6.2)	57.1% (6.5)	3.3%
Pitch deaf	13 F / 3M	21.6 (19-31)	15.4 (2-5)	63.9% (5.7)	81.5% (6.0)	55.6% (7.4)	1.5%
Beat deaf	23 F / 13 M	26.0 (19-39)	17.3 (2-6)	87.8% (7.0)	62.8% (5.1)	84.8% (9.4)	3.3%
Non-Amusics	554 F / 418 M	24.1 (18-40)	16.8 (2-6)	89.2% (6.8)	86.9% (7.33)	87.9% (8.4)	

* 2 = Some undergraduate completed
(students outside of Québec who have completed high school – grade 12)
3 = Some undergraduate completed
(Québec students who have completed Cégep education)
4 = Bachelors completed
5 = Masters completed
6 = Doctorate completed

Table 3: Percentage of Responses (Proportion of Participants) to Questions Relevant for the Identification of Amusic Individuals.

	Pitch memory (n = 35)	Pitch perception (n = 36)	Pitch deaf (n = 16)	Beat deaf (n = 36)	Non-Amusics (n = 972)
Can rarely recognize a very familiar tune without the help of lyrics	28.6% (10/35)	33.3% (12/36)	37.5% (6/16)	8.3% (3/36)	6.1% (59/972)
Unable to detect when someone sings out-of-tune	22.9% (8/35)	41.7% (15/36)	43.8% (7/16)	8.3% (3/36)	5.7% (55/972)
Can rarely recognize out-of-tune notes	40.0% (14/35)	52.8% (19/36)	81.3% (13/16)	11.1% (4/36)	11.9% (116/972)
Sings out of tune	80% (28/35)	86.1% (31/36)	100% (16/16)	52.8 (19/36)	44.1% (429/972)

Table 4: Percentage of Responses (Proportion of Participants) to Questions Relevant for Rhythmic Ability.

	Pitch memory (n = 35)	Pitch perception (n = 36)	Pitch deaf (n = 16)	Beat deaf (n = 36)	Non-Amusics (n = 972)
Does not dance	37.1% (13/35)	44.4% (16/36)	43.8% (7/16)	41.7% (15/36)	35% (343/972)
Does not think they are a good dancer	31.4% (11/35)	38.9% (14/36)	62.5% (10/16)	30.6% (11/36)	34% (329/972)
Cannot dance	22.9% (8/35)	36.1% (13/36)	56.3% (9/16)	30.6% (11/36)	27.5% (268/972)
I cannot follow a musical rhythm	31.4% (11/35)	22.2% (8/36)	43.8% (7/16)	19.4% (7/36)	7.6% (74/972)

Table 5: Percentage of Responses (Proportion of Participants) to Questions about Musical Education

	Pitch memory (n = 35)	Pitch perception (n = 36)	Pitch deaf (n = 16)	Beat deaf (n = 36)	Non-Amusics (n = 972)
School	54.3% (19/35)	61.1% (22/36)	75% (12/16)	77.8% (28/36)	62.1% (604/972)
Optional	28.6% (10/35)	16.7% (6/36)	25% (4/16)	25% (9/36)	42.1% (409/972)
Private	34.3% (12/35)	13.8% (5/36)	25% (4/16)	47.2% (17/36)	51.5% (501/972)
Conservatory	2.9% (1/35)	8.3% (3/36)	0% (0/16)	5.6% (2/36)	11.9% (116/972)
Self-taught	17.1% (6/35)	8.3% (3/36)	6.3% (1/16)	52.8% (19/36)	42.4% (412/972)

Table 6: Family aggregation data

a) Mothers with musical difficulties

	Yes	No	Don't know	Unreported
Pitch memory (n = 35)	14.3% (5/35)	45.7% (16/35)	37.1% (13/35)	2.9% (1/35)
Pitch perception (n = 36)	5.6% (2/36)	33.3% (12/36)	55.6% (20/36)	5.6% (2/36)
Pitch deaf (n = 16)	6.3% (1/16)	25% (4/16)	68.8% (11/16)	0
Beat deaf (n = 36)	8.3% (3/36)	63.8% (23/36)	25% (9/36)	2.8% (1/36)
Non-amusic (n = 972)	13.6% (132/972)	56.8% (552/972)	26.0% (252/972)	3.7% (36/972)

b) Fathers with musical difficulties

	Yes	No	Don't know	Unreported
Pitch memory (n = 35)	14.3% (5/35)	54.3% (19/35)	28.6% (10/35)	2.9% (1/35)
Pitch perception (n = 36)	11.1% (4/36)	27.8% (10/36)	55.6% (20/36)	5.6% (2/36)
Pitch deaf (n = 16)	18.8 (3/16)	25% (4/16)	56.3% (9/16)	0
Beat deaf (n = 36)	13.8% (5/36)	66.7% (24/36)	16.7% (6/36)	2.8% (1/36)
Non-amusic (n = 972)	13.8% (134/972)	52.0% (505/972)	30.7% (298/972)	3.6% (35/972)

c) At least one sibling with musical difficulties

	Pitch memory (n = 35)	Pitch perception (n = 36)	Pitch deaf (n = 16)	Beat deaf (n = 36)	Non-Amusics (n = 972)
Sibling with difficulty	20.0% (7/35)	22.2% (8/36)	25% (4/16)	8.3% (3/36)	16.2% (157/972)

Table 7: Music Habits of Amusics and Non-Amusics

	Amusics (n = 126)	Non-Amusics (n = 972)
Frequently listen to music intentionally	80.2% (101/126)	89.1% (866/972)
Frequently sing in private	47.6% (60/126)	64.7% (629/972)
Frequently sing in public	10.3% (13/126)	19.8% (192/972)

Table 8: Music Habits of Amusics by Type of Amusia and Non-Amusics

	Listen to music		Sing in private		Sing in public	
	Rarely	Frequently	Rarely	Frequently	Rarely	Frequently
Pitch memory (n = 35)	20% (7/35)	71.4% (25/35)	48.8% (17/35)	42.9% (15/35)	85.7% (30/35)	5.7% (2/35)
Pitch perception (n = 36)	16.7% (6/36)	77.8% (28/36)	58.3% (21/36)	38.9% (14/36)	88.9% (32/36)	8.3% (3/36)
Pitch deaf (n = 16)	12.5% (2/16)	81.3% (13/16)	56.3% (9/16)	43.8% (7/16)	93.8% (15/16)	6.3% (1/16)
Beat deaf (n = 36)	11.1% (4/36)	88.9% (32/36)	38.9% (14/36)	61.1% (22/36)	83.3% (30/36)	16.7% (6/36)
Non-amusic (n = 972)	8.2% (80/972)	89.1% (866/972)	32.8% (319/972)	64.7% (629/972)	79.3% (771/972)	19.8% (192/972)

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