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Dominique T. Vuvan, Marilia Nunes-Silva, and Isabelle Peretz

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Dominique T. Vuvan

International Laboratory for Brain, Music, and Sound Research (BRAMS)

Université de Montréal

and Centre for Research on Brain, Language, and Music (CRBLM)

Marilia Nunes-Silva

Developmental Neuropsychology Laboratory, Universidade Federal de Minas Gerais

Isabelle Peretz

International Laboratory for Brain, Music, and Sound Research (BRAMS)

Université de Montréal

and Centre for Research on Brain, Language, and Music (CRBLM)

Author Note

Dominique T. Vuvan, International Laboratory for Brain, Music, and Sound Research, Department of Psychology, Université de Montréal, and Centre for Research on Brain, Language, and Music.

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Correspondence concerning this article should be addressed to Dominique T. Vuvan, BRAMS, Pavillon 1420 Mont Royal, FAS – Département de psychologie, CP 6128, succ. Centre Ville, Montréal, QC, H3C 3J7. Contact: d.vuvan@gmail.com

Abstract

A major theme driving research in congenital amusia is related to the modularity of this musical disorder, with two possible sources of the amusic pitch perception deficit. The first possibility is that the amusic deficit is due to a broad disorder of acoustic pitch processing that has the effect of disrupting downstream musical pitch processing, and the second is that amusia is specific to a musical pitch processing module. To interrogate these hypotheses, we performed a meta-analysis on two types of effect sizes contained within 42 studies in the amusia literature: the performance gap between amusics and controls on tasks of pitch discrimination, broadly defined, and the correlation between specifically acoustic pitch perception and musical pitch perception. To augment the correlation database, we also calculated this correlation using data from 106 participants tested by our own research group. We found strong evidence for the acoustic account of amusia. The magnitude of the performance gap was moderated by the size of pitch change, but not by whether the stimuli were composed of tones or speech. Furthermore, there was a significant correlation between an individual's acoustic and musical pitch perception. However, individual cases show a double dissociation between acoustic and musical processing, which suggests that although most amusic cases are probably explainable by an acoustic deficit, there is heterogeneity within the disorder. Finally, we found that tonal language fluency does not influence the performance gap between amusics and controls, and that there was no evidence that amusics fare worse with pitch direction tasks than pitch discrimination tasks. These results constitute a quantitative review of the current literature of congenital amusia, and suggest several new directions for research, including the experimental induction of amusic behaviour through transcranial magnetic stimulation and the systematic exploration of the developmental trajectory of this disorder.

Keywords: congenital amusia, modularity, pitch discrimination, music cognition

Meta-analytic evidence for the non-modularity of pitch processing in congenital amusia

Congenital amusia is defined as a lifelong deficit in melody perception and production that cannot be explained by hearing loss, brain damage, intellectual deficiencies, or lack of music exposure (Peretz, 2001). Its first description was made possible by the use of a standardized tool, the Montreal Battery for the Evaluation of Amusia (MBEA; Peretz, Champod, & Hyde, 2003) which provided the music neuroscience community with an objective, empirically-derived method by which congenital amusia (henceforth referred to simply as “amusia”) could be diagnosed. In the decade since, this area of research has proliferated, with nearly 300 published studies citing the MBEA.

One of the major themes driving research on amusia is related to the specificity of the musical disorder. Peretz and Coltheart (2003) proposed a modular model of music processing (Figure 1), which describes three pitch organization (tonal encoding, interval analysis, and contour analysis) and two temporal organization (rhythm analysis and meter analysis) components essential to the processing of music. In this model, the tonal encoding of pitch is conceived as the core processing component that is specific to music and hence, modular (Peretz, 2006). One of the MBEA tests, the scale test, assesses the tonal encoding of pitch (“tonal” pitch will henceforth be referred to as “musical” pitch). Most amusics fail to obtain normal scores on this test, and thus the scale test is often used as one of the key diagnostic criteria for amusia (e.g., Hyde & Peretz, 2004; Liu, Jiang, et al., 2012; Loui & Schlaug, 2012; Williamson & Stewart, 2010).

However, the origin of the deficit in musical encoding of pitch seems to arise from a non-modular, lower level of pitch processing. Specifically, the musical deficit is associated to an acoustical deficit in fine-grained pitch discrimination (see Peretz et al., 2002 for an initial report).

In other words, the amusic deficit has been proposed to lie in the acoustic analysis of sounds, and since music processing lies downstream from acoustic processing and makes high demands on fine-grained pitch resolution, amusics manifest a musical impairment. The literature has since been converging towards the finding that amusics display significantly worse pitch discrimination performance as compared to controls (e.g., Hutchins, Gosselin, & Peretz, 2010; Jiang et al., 2012; Jiang, Hamm, Lim, Kirk, & Yang, 2010; Liu et al., 2013; Liu, Jiang, et al., 2012; Liu, Patel, Fourcin, & Stewart, 2010; Nan, Sun, & Peretz, 2010; Omigie & Stewart, 2011; Patel, Wong, Foxton, Lochy, & Peretz, 2008; Tillmann, Burnham, et al., 2011; Tillmann, Rusconi, et al., 2011), bearing out Peretz et al.'s (2002) hypothesis. Indeed, in addition to poor performance on the MBEA, a pitch discrimination deficit has become a diagnostic benchmark for the identification of new amusic cases.

Interestingly, there is evidence for the music specificity, and hence modularity of the disorder. For instance, amusics seem to perceive speech normally, and they can recognize and sing songs through lyrics but not the tune (Ayotte, Peretz, & Hyde, 2002; Dalla Bella, Giguère, & Peretz, 2009; Tremblay-Champoux, Dalla Bella, Phillips-Silver, Lebrun, & Peretz, 2010). However, amusics do show an impairment of pitch perception in speech (Hutchins, Gosselin, et al., 2010; Liu et al., 2010; Patel et al., 2008; Tillmann, Rusconi, et al., 2011), but as Peretz (2013) notes, this deficit is mild relative to their impairment with musical materials. Assuming modular separation between music and speech, one could argue that, because of their musical pitch deficit, amusics use speech mechanisms to process music, but incur a cost, since these mechanisms are not adapted to musical input (Tillmann, Rusconi, et al., 2011). Thus, amusia is a musical disorder that results from a pitch deficit that may (or may not) be acoustic in origin.

One useful way to adjudicate these competing accounts would be to take advantage of the large number of studies that have been carried out on amusics over the last decade. As discussed above, many researchers have included behavioural measures of pitch discrimination in their studies as an ancillary diagnostic (to the MBEA) for amusic cases. We can thus use a meta-analytic approach to pitch discrimination measures in the amusia literature to determine whether the data favour an acoustic or musical locus for the amusic pitch deficit. Meta-analysis has been argued to contribute to the development of knowledge in a field by providing an integrated overview of past findings, as well as stimulating new research efforts (Chan & Arvey, 2012). Furthermore, there is a current movement in psychology towards encouraging replication, in light of the discovery of the low replicability in the most-cited results in psychology (see Special Section on Replicability in Psychological Science: A Crisis of Confidence?, 2012). A meta-analysis is one way to assess the degree to which a particular effect has been consistently replicated in the existing literature.

For this quantitative review, we considered all tasks in which pitch was the critical dimension cuing participant judgments, including both acoustic and non-acoustic (speech and music) tasks; these tasks are summarized in Table 1. Several aspects of the literature can be assessed for evidence for and against pitch modularity in amusia: (1) Whether or not the difference in performance between amusics and controls, henceforth referred to as the “performance gap”, is moderated by variations in acoustic difficulty; (2) Whether the performance gap depends on the stimuli being composed of tones versus speech; (3) Whether acoustic pitch performance is correlated with musical pitch performance; and (4) Whether individual cases exist in which acoustic and musical deficits are dissociated.

If the amusic deficit is acoustic in origin, variations in the acoustic difficulty of the pitch discrimination task across studies should moderate the performance gap. For instance, Hyde and Peretz (2004) found that amusics had trouble distinguishing pitch changes of 100 cents (1 semitone), whereas controls performed close to ceiling down to changes of 25 cents. Accordingly, the acoustic account suggests that across studies we should observe a linear relation between size of pitch change and the performance gap, such that small pitch changes are associated with larger performance gaps between amusics and controls. An acoustic origin also suggests that the performance gap should be constant across stimuli composed of tones and speech, because it is the size of the acoustic pitch change, not the domain in which it is presented that matters. Furthermore, the severity of the acoustic deficit should be correlated with the severity of the resultant musical deficit, because of the putative causal link from acoustic to musical pitch processing. Finally, if the origin of the amusic deficit is truly acoustic, scrutiny of individual data should reveal that every amusic case (i.e., individual with low MBEA scale test score), also performs poorly in an acoustical perception task.

The alternative hypothesis is that the amusic pitch deficit is specific to the tonal encoding module (Peretz & Coltheart, 2003), and thus to music. This modular account does not advocate for any specific effect of acoustic difficulty, except that there should be a clear performance gap at 100 cents, the smallest categorical pitch interval in Western music, as found by Hyde and Peretz (2004). However, it does predict that the size of the performance gap should be larger for tones than speech. Moreover, acoustic pitch discrimination should be dissociable from musical pitch discrimination. Thus, we should observe weak evidence for a correlation between acoustic and musical pitch performance, and consequently, we should be able to identify individuals who

have poor acoustical pitch perception but who are not amusic, and conversely, amusic individuals who demonstrate normal acoustical pitch perception.

In order to test these hypotheses, we investigated patterns of effect sizes across studies in two different ways. For the first analysis, we performed a meta-analytic review of the literature on amusia, following the guidelines of Johnson and Eagly (2000) and Rosenthal and DiMatteo (2001). Here, we considered as effect sizes the amusic-control performance gap on tasks of pitch discrimination, broadly defined (Table 1). For the second analysis, we considered as effect sizes the correlations between performance on specifically acoustical tests of pitch processing (Table 1) and scores on another task of interest, namely the MBEA scale test. Since the individual data required for this latter type of analysis are less commonly reported in the literature, we also included data from the large database (the “Montreal database”) of amusic and control performance collected by our research group over the last decade.

For the meta-analysis, we created several vectors that coded, for each effect in the database, factors that might systematically moderate effect size across studies. First, in order to assess the modularity of the amusic pitch deficit, we coded vectors that contained the size of pitch change used in the task and whether the stimuli were composed of tones or speech. Next, we coded vectors corresponding to certain other factors of interest. Specifically, we coded the various effects according to participant age, the duration of the target tone or syllable, whether the experimental task required detecting a change in pitch or the direction of the change, and whether participants spoke a tonal language.

Age was of interest because pitch discrimination abilities change over the lifespan, due to changes in both peripheral and central mechanisms of hearing (Gordon-Salant, Frisina, Popper, & Fay, 2010). This phenomenon should lead to a higher prevalence of acoustical pitch disorders

with no associated musical disorder in older participants, in support of the modularity hypothesis. Furthermore, musical training is known to alter the developmental trajectory of acoustical abilities (e.g., Parbery-Clark, Anderson, Hittner, & Kraus, 2012; Parbery-Clark, Strait, Anderson, Hittner, & Kraus, 2011; Zendel & Alain, 2012). Given the altered musical experience of amusics, it is possible that the pitch systems of amusics and controls develop differently throughout the lifespan. In this case, the size of the pitch discrimination performance gap may change depending upon participant age.

Target duration was of interest because of the finding that amusics perform worse than controls on pitch memory tasks such as tone span (Albouy, Mattout, et al., 2013; Albouy, Schulze, Caclin, & Tillmann, 2013; Tillmann, Schulze, & Foxton, 2009; Williamson, McDonald, Deutsch, Griffiths, & Stewart, 2010; Williamson & Stewart, 2010). This finding might be interpreted as symptomatic of amusics' failure to adequately encode incoming pitch information, making it difficult to form pitch traces in memory. If this account holds, then stimuli with longer targets will support better pitch encoding than stimuli with shorter targets, and the performance gap might be ameliorated when target duration is longer.

Pitch change vs. direction was of interest because some authors have found that amusics' pitch direction deficit was greater in magnitude than their pitch change detection deficit, regardless of direction (Foxton, Dean, Gee, Peretz, & Griffiths, 2004; Liu et al., 2010). This meta-analysis is thus a good opportunity to assess the validity of this claim over many studies.

Lastly, whether participants spoke a tonal language is of interest because tonal languages use subtle pitch contrasts to convey meaning (Yip, 2002). Existing studies indicate that the prevalence of amusia is similar across tonal and non-tonal language speakers (approximately 3-5%; Nan et al., 2010; Wong et al., 2012), providing some evidence for the modularity account.

On the contrary, some research has demonstrated speech comprehension difficulties in amusic tonal language speakers, indicating that the amusic pitch processing deficit is not specific to music. For instance, Jiang et al. (2012) showed that Mandarin-speaking amusics are impaired in a speech comprehension task in which they were asked to classify pitch-based prosody as appropriate or inappropriate, with an individual's performance being significantly correlated with their aggregated score for the pitch-based tests of the MBEA. Additionally, amusic participants did not exhibit the same evoked responses in the N100 and P600 components as controls when listening to trials with inappropriate prosody. Furthermore, a recent study found that this speech comprehension difficulty remains when pitch is flattened (Liu, Jiang, Wang, Xu, & Patel, 2015). However, other work reveals that Mandarin-speaking amusics perform equally to controls given multiple acoustic cues to meaning (Liu, Jiang, et al., 2012), indicating that speech processing is only significantly impaired in tone language-speaking amusics when pitch is the task-relevant cue. On one hand, this finding provides some support for shared pitch processing between music and speech in amusia. On the other hand, this finding indicates that whatever the nature of the amusic pitch processing deficit, it only functionally affects music processing whereas speech processing is preserved due to cue redundancy. A meta-analytic approach thus provides the ability to adjudicate between these two views via a comparison of pitch discrimination thresholds between tonal language and non-tonal language speakers, which has not been explicitly assessed in previous research.

The comparison of tonal language speakers and non-speakers also impinges upon the question of whether amusics can improve through pitch discrimination training. Tonal language learning can be thought of as an indirect form of this training, as speakers learn to focus on small changes in pitch due to their semantic importance; in amusic tonal language speakers, this speech

skill might then be generalized to musical contexts, thus somewhat alleviating the amusic pitch deficit. In this view, it is possible that other types of pitch discrimination training would also be effective in improving the amusic pitch deficit. For instance, an ongoing study in our lab seeks to train amusic teenagers to better discriminate pitch through guitar lessons (Mignault Goulet et al., in preparation).

Methods

Search Procedure

The literature was searched through November 2013. Google Scholar was used exclusively as a search database, following recent research that demonstrated that the search results for previously published systematic reviews (which used GS, in addition to PubMed, Cochrane, Dissertation Abstracts International, and a variety of other sources), were 100% covered by Google Scholar (Gehanno, Rollin, & Darmoni, 2013). Searches were executed using the following terms: *amusia*, *auditory agnosia*, *dysmelodia*, *melody deafness*, *note blindness*, *pitch deafness*, *poor pitch singing*, *tone deafness*.

Inclusion and Exclusion Criteria for Database

Prior to the development of the MBEA, formal assessments of musical ability existed. However, these tests were either not informed by recent empirical neuropsychological work (e.g., Seashore Measures of Musical Talent; Seashore, 1939) or contaminated by prior knowledge of the test material (e.g., Distorted Tunes Test; Kalmus & Fry, 1980)¹. Thus, the first

¹ The DTT is no longer considered an appropriate diagnostic tool for amusia. In addition to the issue of long-term memory for familiar melodies, the DTT does not include any additional tests to control for more generalized deficits that would lead to poor performance on the DTT task itself, such as deficits in IQ, attention, working memory, or peripheral hearing processes. Therefore, individuals who fail the DTT may be congenitally amusic, but they might also suffer from any of many possible unrelated disorders.

inclusion criterion was the use of the MBEA for diagnosis of amusic subjects. Application of this criterion formed an initial database of 117 unique articles. Furthermore, studies were required to: (a) study congenital, not acquired, amusia; (b) be published in English; (c) contain data for a sample of amusics and a sample of controls, the performance for which can be compared via the performance gap (i.e., no case studies); (d) report behavioural measures of pitch discrimination, broadly defined (Table 1). The application of all selection criteria resulted in the inclusion of 43 unique articles. One of these articles (Liu et al., 2013) was dropped from the database because it reported the same pitch threshold data (for the same set of participants) as another article that was already included (Liu, Jiang, et al., 2012).

Performance Gap Analysis

Quantifying the effect sizes. For the performance gap analysis, the effect size was a quantification of the differential performance of amusics and controls on the pitch discrimination task of interest. For each result in each study, the effect size was extracted (t , F , or z), and then converted to the r metric for its ease of interpretation (Rosenthal & DiMatteo, 2001). These r values were then adjusted for bias using the approximation of the population effect size G (Johnson & Eagly, 2000), and coded as positive if amusics performed worse, and negative if amusics performed better than controls on the task. Our performance gap database comprised 88 effect sizes contained within 42 studies (Table 1).

Coding of Moderators. The creation of vectors coding for moderating factors was performed by the first two authors. Reliabilities were then established on 35% of the dataset; the intercoder reliability for each moderator (calculated as the percent of agreement between the two coders on that 35% of the dataset) is reported below in the paragraph describing that moderator. The two coders were not blind to the data source due to the necessity of close reading to

determine the value of the various moderators. Disagreements between coders were all due to calculation errors (e.g., range/average of pitch change; average number of syllables; average sentence length) by one or both coders, and were resolved by consensus following discussion, re-reading of the article in question, and recalculation. Moderators were coded for each effect size rather than each study; therefore, studies that included multiple effect sizes received a distinct code for each moderator, for each effect size. Descriptive statistics for these moderators are shown in Tables 2 and 3 (continuous and dichotomous moderators, respectively).

Size of pitch change was a continuous variable coded in cents (percentage of a semitone, the smallest pitch change employed in music). Cents are logarithmically related to physical frequency (e.g., Hertz), as is the human perception of pitch height. For studies that supplied only the range of pitch changes (i.e., minimum and maximum), the centre of the range was calculated. For instances where multiple pitch change sizes were collapsed into one result, the average was calculated. Notably, size of pitch change was only meaningful for tasks in which the pitch changes were fixed, i.e., the same pitch changes were administered to every participant, amusic or control (58 effect sizes). For a significant proportion of results, adaptive tasks were used to identify an individual pitch change detection threshold for each participant (29 effect sizes). In these latter cases, the performance gap would be measured by the difference in threshold between controls and amusics, and thus is confounded with the size of the pitch change employed for each participant in the study. Thus, fixed and adaptive pitch change studies were considered separately in the latter part of the moderator analysis (see below). The intercoder reliability for this variable was 93%.

Tone vs. speech was a dichotomous variable, with effect sizes being coded according to whether the stimulus was comprised of tones or speech. In certain studies where results were

collapsed across speech and tone stimuli, the stimulus timbre was not given a value. The intercoder reliability for this variable was 100%.

Participant age was a continuous variable coded in years. This was calculated, where possible, as the weighted mean of the average ages of the amusic and control subjects. If a range was provided rather than means, the centre of the range was taken. Finally, in one study (Hutchins, Zarate, Zatorre, Peretz, 2010), where only data for the amusics was provided (but controls were matched for age), the mean of the amusics' ages was used. The intercoder reliability for this variable was 100%.

Target duration was a continuous variable coded in milliseconds (ms). "Target" refers to the critical tone or syllable within a stimulus that contains the pitch change. In studies where target duration was not explicitly given (i.e., many studies using speech), target duration was estimated from the average number of syllables within a stimulus and the average sentence length. The intercoder reliability for target duration was 85%.

Pitch change vs. direction was a dichotomous variable, with effect sizes being coded according to whether the task required the discrimination of a change, irrespective of its direction, or whether direction was key to a correct judgment. The intercoder reliability for this variable was 100%.

Participant language was also a dichotomous variable, with effect sizes being coded according to whether participants in that study spoke a tonal language or not. The intercoder reliability for this variable was 100%.

Assessing hierarchical dependencies. Before building a meta-analytic model for the data, a methodological decision had to be made concerning the appropriate unit of analysis. There are a small number of research groups producing the majority of the studies in this

domain, and these studies often draw repeatedly from a small pool of participants. Additionally, multiple effect sizes are often reported within the same study. Thus, there seem to be hierarchical dependencies built into these data (effect sizes nested within studies, studies nested within research groups). In order to determine whether this hierarchy should be modeled in our meta-analysis, it was important to assess the quantitative reality of these dependencies. To this end, each effect size was coded according to two variables: the study in which the result was reported, and the research group that published the study. The following steps were executed for both the performance gap and correlation data.

A study was defined as an experiment or series of experiments that was run on a single set of participants. In nearly all cases, this corresponded to one study per published article. In the one exception (Patel et al., 2008), the authors reported two studies. With regard to the “research group” variable, all authors of the 42 studies included in our database could be reduced into five research groups, associated with C. Jiang, I. Peretz, G. Schlaug, L. Stewart, and B. Tillmann. Thus, any study with one of these individuals as authors was coded as coming from that research group. In the event that more than one of these individuals appeared as authors on a single paper, the paper was attributed to the research group whose lab tested the participants. For example, Tillmann, Rusconi, et al. (2011) was coded as coming from the Peretz group, and Liu, Jiang, et al. (2012) was coded as coming from the Jiang group. As with the moderators, reliabilities were then established on 35% of the dataset; intercoder reliability was 100% for both study and research group variables.

For each performance gap and correlation analysis reported in the results section, a baseline (no predictors) hierarchical linear model (effect sizes nested within studies nested within research groups) was considered. Within these nested models, the variance for each effect size

was weighted by the inverse of the corresponding sample size ($1/N$; Johnson & Eagly, 2000). Interestingly, the intraclass correlations obtained for these hierarchical models were uniformly low ($< .10$) and the likelihood ratio test comparing the hierarchical model to the non-nested model (in which each effect size was treated as an independent observation) was non-significant ($p > .05$) for all analyses. Our effect sizes could thus be considered statistically independent from one another, and were modeled as such for all performance gap analyses.

Analysis procedure. We performed a two-stage analysis of the performance gap effect sizes (Johnson & Eagly, 2000), starting with the summary of the effect sizes at the study level (i.e., multiple effect sizes within each study were collapsed), and then subsequently considered these effect sizes as independent for the analysis of the potential moderators. All meta-analytic models were fitted using the metafor package (Viechtbauer, 2010) in R (R Core Team, 2013).

Overall effect size. This analysis was performed at the level of the study. To combine multiple results in the case that a study reported more than one effect size, the G -corrected r values were transformed to Fisher's z scores, a mean was calculated, and then this mean was converted back to an r value (Silver & Dunlap, 1987). Central tendency was estimated using several measures, including unweighted mean r , median r , and weighted mean r (by sample size). In order to estimate the weighted mean r and its associated significance level and confidence interval, all r values were entered into a random effects model using the *rma* function with restricted maximum-likelihood (REML) estimation of residual heterogeneity from the metafor package. In order to assess the possibility that any of the study effect sizes acted as influential outliers in this analysis, Cook's distances were calculated for each study using the *influence* function of the metafor package (with a threshold of $d > 1$).

Moderators. These analyses were performed at the level of the individual effect sizes. First, a baseline random effects model (no moderators) was fitted using the *rma* function with REML estimation, and Cook's distances were calculated to eliminate outliers. This function also estimates $\hat{\tau}^2$ and I^2 , which refer to the amount of residual heterogeneity and the percentage of that heterogeneity not due to sampling error, respectively. These statistics, in combination with the Q significance test for heterogeneity, can be used to infer the usefulness of moderator analyses, since a significant amount of variability in effect sizes can indicate the explanatory usefulness of moderating factors.

Second, to get a general idea for the predictive value of each moderator in the absence of the others, we fitted simple regressions for each moderator. To do this, we built a mixed effects model using *rma* and REML estimation. In addition to the significance level of the moderator, comparing the heterogeneity statistics from these simple moderator models with the baseline model enabled us to assess each moderator's contribution to effect size.

Third, to get an idea of how the moderators might relate to one another, we built a mixed effects model that predicted all corrected r values from a combination of the moderators that were significantly predictive in the simple models, as well as their interactions. All continuous predictors were mean-centred. Models were estimated separately for effect sizes resulting from fixed and adaptive pitch change methods, since neither size of pitch change nor stimulus timbre could be used as moderators for adaptive pitch studies (for adaptive studies the size of pitch change was not fixed, and the stimuli were always composed of tones).

Correlation Analysis

Quantifying the effect sizes. For the correlation analyses, the effect size of interest was the Pearson r calculated by relating participants' MBEA scale score and acoustic (not musical, or

speech) pitch discrimination performance. Therefore, only the first six pitch discrimination tasks from Table 1 were considered. Where possible, r was taken directly from the article. When raw participant data was provided, r was calculated from these data. These r values were then adjusted for bias with G , as above. When pitch discrimination was measured via a threshold, thus giving r a negative sign if there was a positive correlation between pitch discrimination performance and the MBEA scale score, the r was re-coded with the opposite sign for consistency with other values. Our correlation database was comprised of seven effect sizes contained within four studies.

To augment the small meta-analytic database, the participant scores from the Montreal database (compiled from our research group's data) were used to calculate a similar correlation. Here, pitch discrimination performance was measured via the task used by Hyde and Peretz (2004), consisting of trials containing a sequence of five pure tones. The fourth tone was either identical to the other tones or deviated from the other tones (by 25-200 cents); participants were required to judge whether the fourth tone was the same or different from the other tones. We considered the Montreal data separately from the meta-analytic database because there is partial overlap between the data herein and data presented in some of the published studies from our research group.

Assessing hierarchical dependencies. Just as we assessed the performance gap database for hierarchical dependencies, so too did we assess the correlation database. Each effect size was coded according to study and research group (intercoder reliability = 100%), and then a baseline hierarchical linear model was calculated, weighted for each sample size. Like for the performance gap data, the intraclass correlations for this model was low ($< .10$) and the

likelihood ratio test was not significant ($p > .05$). We thus considered these effect sizes to be statistically independent and modeled them as such.

Analysis procedure. Meta-analysis of the correlations between pitch discrimination and MBEA scale test scores from the literature followed the methods described above for the calculation of overall effect size for the performance gap data. Specifically, central tendency was estimated by unweighted mean r , median r , and weighted mean r (by sample size). In order to estimate the weighted mean r and its associated significance level and confidence interval, all r values were entered into a random effects model using the *rma* function with restricted maximum-likelihood (REML) estimation of residual heterogeneity from the *metafor* package. In order to assess the possibility that any of the study effect sizes acted as influential outliers in this analysis, Cook's distances were calculated for each study using the *influence* function of the *metafor* package (with a threshold of $d > 1$).

Results

Performance Gap: Overall Effect Size

At the level of the study, the mean weighted effect size was $r = 0.61$ ($SE = 0.03$), $p < .0001$. This is a fairly strong effect, with low variability, indicating that controls consistently outperform amusics on pitch-based musical tasks. The unweighted mean and median (0.56 and 0.54, respectively) were slightly lower than the weighted mean, indicating that studies with larger participant samples tended to report higher effect sizes. Figure 2a shows a forest plot of the effect sizes (r bias-corrected with G) by study, and Figure 2b shows the distribution of those effect sizes. The calculation of Cook's distances indicated that no outlying effect sizes significantly influenced the model estimate (all $D < 4N$; Bollen & Jackman, 1985). The Shapiro-

Wilk test indicates that the study-level effect sizes conformed to a normal distribution, $W = 0.99$, $p = .88$.

Performance Gap: Moderator Analysis

Baseline model. At the level of the effect sizes, the mean weighted effect size was $r = 0.55$ ($SE = 0.03$), $p < .0001$. Confirming the overall effect size analysis, the mean effect is quite high, with low variability, with controls consistently outperforming amusics. The unweighted mean and median (0.51 and 0.53, respectively) were slightly lower than the weighted mean, again consistent with the previous analysis. Figure 3a shows a forest plot of all effect sizes, and Figure 3b shows the distribution of those effect sizes. The calculation of Cook's distances indicated that one effect size from Jiang, Lim, Wang, and Hamm (2013), with a value of $r(26) = -0.57$, exceeded Bollen and Jackman's (1985) criterion. Thus, this effect size was excluded from further analysis. The Shapiro-Wilk test indicates that these effect sizes conformed to a normal distribution, $W = 0.99$, $p = .18$.

The estimates of heterogeneity for the random-effects model revealed that the overall amount of heterogeneity was fairly low, $\hat{\tau}^2 = 0.05$, confirming the small estimate of standard error of the weighted mean effect size (0.03). Despite the small variability in effect size, a large proportion of that variability could potentially be accounted for by our hypothesized moderators, $I^2 = 0.89$, $Q(87) = 942.48$, $p < .0001$. Thus, the next analysis was aimed at determining which of these moderators were significant predictors of the variability in effect sizes.

Simple regressions. We used simple regression models to predict effect size from each moderator alone. The moderators tested were, as described above: size of pitch change, tone vs. speech, participant age, target duration, pitch change vs. direction, and participant language. These analyses revealed two moderators of interest. Size of pitch change had a significant impact

on effect size, $b = -0.0002$, $SE = 0.0001$, $p < .0001$, with larger pitch changes corresponding to a smaller performance gap between controls and amusics (Figure 4). Whether or not the participant spoke a tonal language also had a significant effect on the effect size, $b = -0.11$, $SE = 0.05$, $p = .04$, with smaller performance gaps between controls and amusics for studies of tonal language speakers (25 effect sizes, mean $r = 0.43$, $SD = 0.27$) than studies of non-tonal language speakers (63 effect sizes, mean $r = 0.54$, $SD = 0.24$).

None of the other hypothesized moderators (tone vs. speech, participant age, target duration, or pitch change vs. direction) were significantly predictive of effect size in these simple models, all $p > .09$. Particularly of interest is the lack of effect for tone vs. speech, indicating that the average performance gap in studies employing tone stimuli is the same as that in studies employing speech stimuli (mean for tones $r(66) = 0.51$, $SD = 0.27$; mean for speech $r(15) = 0.50$, $SD = 0.16$). This is a key contrast relative to the assessment of the modularity of congenital amusia, and this result supports the idea that the locus of the amusic disorder is in acoustic processes which affect pitch processing in music and speech equally.

Multiple regression for fixed pitch studies. Next, we performed a multiple regression analysis using only fixed pitch studies (for which the size of pitch change variable was meaningful). As suggested by our simple regression analyses, this model included the size of pitch change, participant language, and the interaction between these two variables as predictors of the performance gap. The size of pitch change was a significant of the performance gap, $b = -.0002$, $SE = .00006$, $p = .002$. Neither the main effect of participant language nor the interaction were significant after controlling for the effect of the size of pitch change, all p values $> .21$.

Multiple regression for adaptive pitch studies. Given that the size of the pitch change was not meaningful for adaptive pitch studies, the only valid moderator that could be used as a

predictor for a multiple regression is participant language. Thus, the appropriate model is given by the simple regression analysis using participant language to predict the performance gap, reported above.

Correlations: Meta-analytic Database

The mean weighted effect size for the correlation between the MBEA scale score and the pitch discrimination measure in the study was $r(5) = 0.46$ ($SE = 0.09$), $p < .0001$. This result indicates that acoustic pitch performance significantly predicts musical pitch performance. Interestingly, the weighted mean is a bit higher than the unweighted mean and median (0.42 and 0.35, respectively), indicating possible outliers. Figure 5 shows a forest plot of the effect sizes. The calculation of Cook's distances indicated that one effect size from Tillmann et al. (2009), with a value of $r(18) = 0.81$, exceeded Bollen and Jackman's (1985) criterion. The recalculation of the mean weighted effect size, omitting this effect size, yielded a value of $r(5) = 0.39$ ($SE = 0.07$), $p < .0001$.

Correlations: Montreal Database

We analyzed our database for the relation between musical pitch (MBEA scale score) and acoustic pitch discrimination. For the latter, we considered the hit rate – false alarm rate of a 25 cent change in a five repeating tone sequence; this was the smallest pitch change employed in this task, and therefore most difficult to detect (Hyde & Peretz, 2004). These data include 106 participants, 61 amusics and 44 controls matched in age, education and musical background. In our pool, we also found a correlation between the musical pitch and acoustical pitch score with $r(104) = 0.58$, $p < .0001$. The correlation was still significant when considering amusics only, $r(59) = 0.34$, $p = 0.007$. These data confirmed the results from the meta-analytic database,

indicating that musical pitch abilities are significantly associated with acoustic pitch abilities in the population.

As can be seen in Figure 6, there are cases showing dissociation between musical and acoustic pitch performance. We defined the cutoff for poor MBEA scale performance at 22.73 (two SD below the mean of the controls), and the cutoff for poor pitch change detection performance was set at hit rate – false alarm rate = 57.84 (two SD below the mean of controls in our data). Using these cutoffs, we found 17 cases who were normal at processing musical pitch despite having a deficit in acoustical pitch perception (top left quadrant of Figure 6). Thus, a deficit in acoustic pitch perception does not necessarily lead to a musical disorder. Conversely, there were nine amusic cases who had normal pitch perception (bottom right quadrant, Figure 6), indicating that amusia may occur without an associated pitch deficit. Importantly, it is unlikely that the dissociations we have observed here are due to hearing problems. For the subset of these participants (7 of the 17 participants with an acoustic deficit, 6 of the 9 participants with a musical deficit) who had undergone audiometric testing, all had normal pure tone thresholds, standardized for their age (ISO1999 standard, 2013).

Discussion

This quantitative review has demonstrated the reality of the amusic pitch discrimination deficit. The size of this effect, as quantified by the performance gap between amusics and controls, is robust, large, and significant. Additionally, our analyses enabled us to assess the extent to which amusia is specific to the processing of musical pitch, or affects the more general processing of acoustic pitch.

Recall that we investigated four different sources of evidence to determine the cognitive mechanism underlying the pitch discrimination deficit observed in amusia. The first three

sources of evidence found in favour of the long-hypothesized model, wherein an acoustic deficit leads to the downstream malfunction of tonal pitch processing (Peretz et al., 2002; Peretz & Hyde, 2003). Specifically, we found that the size of the pitch change employed in a study, which is an acoustic variable, moderated the effect size. Furthermore, we found no significant moderating effect of tone vs. speech stimuli, which indicates that the amusic deficit affects the processing of pitch in both tones and speech. This is evidence that the locus of this deficit is in acoustic processing, which is upstream from language and music specific modules. Third, we observed a significant correlation between measurements of acoustic functioning (i.e., pitch discrimination) and measurements of musical functioning (i.e., MBEA scale subtest). This is evidence that the extent to which an individual's acoustic pitch processing is compromised is related to the extent to which that individual's musical pitch processing is also compromised, indicating a link between acoustic and musical processing in amusia. Thus, the pitch discrimination deficit observed in amusia is located at both acoustic and musical levels. This is evidence against the modularity of the disorder and suggests that the acoustic account, whereby a general acoustic pitch deficit is responsible for the musical pitch deficit in amusia, is correct.

However, one source of evidence does speak against the conclusion that the amusic deficit is acoustic in nature. Namely, scrutiny of individual cases in our database revealed a double dissociation between acoustic and musical processing. We observed nine cases with poor musical performance alongside normal acoustic performance, and 17 cases with normal musical performance alongside poor acoustic performance out of our database of 106 participants.

The nine amusic cases with normal pitch discrimination suggest that it is possible for an individual to demonstrate the amusic deficit without an acoustic deficit. However, the relation between acoustic and musical pitch abilities in these cases may have changed over the lifespan,

especially because amusia is a developmental disorder. For instance, these participants may have had an acoustic deficit in infancy which impacted their early musical development during a sensitive period (e.g., Habib & Besson, 2009) and caused their musical disorder. At the same time, their acoustic pitch discrimination abilities may have subsequently improved to a normal level due to prolonged experience with pitch in other domains such as speech. At the very least, these cases argue for the existence of heterogeneous phenotypes in the amusic population. It is likely that the majority of amusic cases present an acoustic deficit, but there clearly exist cases who present musical difficulties in the absence of acoustic problems. It is important to emphasize that the core deficit defining amusia is the musical deficit, regardless of whether this deficit is modular.

Additionally, the 17 cases with poor pitch discrimination in the absence of a musical disorder indicate that it is possible to have an acoustic deficit without a resulting musical deficit. Moreover, this acoustic pitch deficit is not caused by general hearing difficulties, as measured by pure tone audiometry. There is a possibility that this is the result of the effects of aging on hearing abilities; the average age of these 17 cases is 58.6 ($SD = 12.4$), with only two of these cases below the age of 50. Although the participants who underwent audiometric testing all had normal pure tone thresholds, these thresholds do not account for all the processing, both peripheral and central, that is undertaken during the acoustic pitch discrimination task. Thus, as acoustic abilities degrade with age (Gordon-Salant et al., 2010), musical perception could ostensibly remain, aided by top-down processing of extra-acoustic cues. However, the dissociation between acoustic and musical processing presents a parsimonious explanation for these cases.

Confirming the nature of the relation between acoustic and musical processing in congenital amusia requires direct experimental study. A potentially fruitful approach would be to use transcranial magnetic stimulation (TMS) in non-amusic participants to simulate the amusic deficit. Amusia is characterized by atypical electrical brain responses during pitch discrimination tasks; in particular, amusics do not exhibit the P300 component in an acoustic pitch discrimination task (Peretz, Brattico, & Tervaniemi, 2005). Various studies have converged in identifying the right inferior frontal cortex (BA47), right auditory cortex (BA22), and the arcuate fasciculus as potential sources of these abnormal electrical responses (Hyde, Zatorre, Griffiths, Lerch, & Peretz, 2006; Hyde, Zatorre, & Peretz, 2011; Loui, Alsop, & Schlaug, 2009), and recent evidence suggests that BA47 is responsible for amusics' abnormal P300 (Albouy, Schulze, et al., 2013). The centrality of these cortical mechanisms to amusia is supported by recent evidence that amusics show intact brainstem encoding of speech and music, as measured by the frequency following response (Liu, Maggu, Lau, & Wong, 2015). Therefore, TMS could be used to mimic amusic brain function in non-amusic participants by creating a transient lesion in BA47. The successful simulation of the amusic deficit in these participants would provide compelling evidence for a causal link between acoustic and musical pitch in the amusic disorder.

In addition to the analysis targeting questions of modularity in amusia, we also assessed the moderating influence of some additional variables of interest on the performance gap. One particularly interesting factor was whether participant spoke a tonal language. A simple regression model indicated a role for participant language, with studies of tonal language speakers reporting a smaller performance gap between amusics and controls. However, when the size of pitch change was controlled, participant language was no longer a significant predictor, nor did it interact significantly with the size of pitch change. This suggests that either participant

language is not a factor in the amusic deficit, or that the effect is too subtle to detect with our analysis. To the latter point, although amusia has been shown to be highly heritable and therefore dependent upon genetics (Peretz, Cummings, & Dubé, 2007), the developmental trajectory of amusia, its interaction with language learning in children and adolescents, and thus the possible effect of experience on the severity of this disorder, is poorly understood. The vast majority of the literature has studied adult amusics, with the notable exception of two studies (Lebrun, Moreau, McNally-Gagnon, Mignault Goulet, & Peretz, 2012; Mignault Goulet, Moreau, Robitaille, & Peretz, 2012). Interestingly, of all the effect sizes included in this meta-analysis, the maximal performance gap ($G = .95$) was observed for the detection of a 25 cent pitch change by the adolescent participants tested by Mignault Goulet et al. (2012). Thus, work in this vein has continued in our research group as well as others. Tonal language speakers receive lifelong implicit pitch discrimination training in a speech context, and this may alleviate the impact of the amusic disorder. This idea advocates for the importance of studies directed at pitch acuity training in amusic participants, with the possibility that the pitch deficit in amusia could be improved through training; several studies addressing this issue are in progress in our lab.

Lastly, we found that target duration did not have a significant impact upon the control-amusic performance gap, nor did pitch change vs. direction. Turning first to target duration, our meta-analytic database contained durations with a mean of 377 ms and a range from 100 to 850 ms. Intriguingly, this range overlaps considerably with the profile of pitch durations used in music (280 ± 291 ms; Patel, 2013). However, there is a large amount of variation in terms of duration in our database, and notably, some studies used very long durations (850 ms) that are less frequent in musical melodies. Given the inclusion of such an extensive range of pitch

durations, this result constitutes strong evidence for the lack of effect of target duration on the performance gap.

The lack of effect for pitch change vs. direction is intriguing, as some individual studies have reported that amusics perform significantly worse on direction tasks than on change detection tasks (e.g., Omigie & Stewart, 2011; Williamson, Cocchini, & Stewart, 2011). One intriguing reason that direction might be more difficult for amusic participants is that the detection of the direction of a pitch change is more explicit a task than the simply noticing that there is a change. Given that amusia has been characterized as a deficit of awareness (Paquette, Goulet, & Rothermich, 2013; Peretz, Brattico, Järvenpää, & Tervaniemi, 2009), the level of explicit awareness of pitch information required for the task may play an important role. However, many studies have reported mixed results regarding whether amusics find direction tasks more difficult than detection tasks (Liu et al., 2013; Liu, Jiang, et al., 2012; Liu et al., 2010; Williamson, Liu, Peryer, Grierson, & Stewart, 2012), and the weight of the evidence here indicates that amusics have equal difficulty with change detection and direction tasks. Additionally, all the tasks assessed by our meta-analysis were explicit ones, and it may be that a small difference in degree of explicitness is not enough to elicit effect size differences between change detection and direction tasks across studies.

In summary, this quantitative review has confirmed the reality of the amusic pitch discrimination deficit, confirmed the acoustic locus of the amusic deficit (albeit with a caveat concerning heterogeneity in the amusic population), and suggested some fascinating avenues for future research. It is likely that this area of study will continue to be productive, as congenital amusia constitutes a natural experiment that offers a window to the many important questions of cognitive neuropsychology. These questions include the investigation of processing modularity

between music and speech, which was a focus of this study (see also Mantell & Pfordresher, 2013 for a recent discussion), and the search for etiological similarities between amusia and other neurodevelopmental disorders such as dyslexia, which like amusia, may result from the impact of a sensory deficit upon cognition (Ramus, 2003).

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Table 1

Pitch discrimination tasks included in meta-analytic database (88 effect sizes)

Acoustic Tasks			
Task	Description	Example	Number of Effect Sizes
Pitch matching	Use a dial to match a tone to a target	Anderson et al. (2012)	1
Pure tone pitch change detection	Determine whether one tone in a set differs from the others	Hyde & Peretz (2004)	10
Pure tone pitch change identification	Determine which of the tones is different from the others	Liu et al. (2010)	15
Pure tone pitch change discrimination	Determine whether two sets of tones are the same or different	Gosselin et al. (2009)	11
Pure tone pitch direction identification	Determine whether the pitch change is up or down	Loui et al. (2008)	16
Pure tone pitch direction discrimination	Determine whether the first two or the last two tone pairs have the same pitch change direction	Jiang et al. (2013)	1
Non-Acoustic Tasks			
Task	Description	Example	Number of Effect Sizes
Melodic pitch judgment	Determine whether the melody contained a wrong note	Peretz et al. (2008)	6
Speech intonation (or complex tone analogue) identification	Determine whether the sentence is a statement or a question	Liu et al. (2010)	10
Speech intonation (or complex tone analogue) discrimination	Determine whether the the intonation of the second sentence is the same as the first	Patel et al. (2008)	16
Effect size amalgamated across more than one pitch discrimination task		Foxton et al. (2004)	2

Note. Studies appearing in the Example column contained at least one effect size corresponding to the Task in that row. The contents of the Number of Effect Sizes column indicate the total number of effect sizes in the database corresponding to the Task in that row.

Table 2

Continuous moderators of the control-amusic performance gap

Moderator	Mean	SD	Median	Range
Size of pitch change (cents)	566	503	400	[25, 1970]
Participant age (years)	42	15	48	[12, 66]
Target duration (ms)	377	204	350	[100, 850]

Table 3

Dichotomous moderators of the control-amusic performance gap

Moderator	Coding Scheme	Number “0”	Number “1”
Tone vs. speech	0 = tone; 1 = speech	68	17
Pitch change vs. direction	0 = change; 1 = direction	63	25
Participant language	0 = non-tonal; 1 = tonal	63	27

Figure Captions

Figure 1. A modular model of music processing, reproduced with permission from Peretz & Coltheart (2003). What appears in green was considered music-specific.

Figure 2. (a) Forest plot and (b) histogram showing performance gap effect sizes (G -corrected r) by study.

Figure 3. (a) Forest plot and (b) histogram showing all performance gap effect sizes (G -corrected r).

Figure 4. The effect of size of pitch change on the performance gap between controls and amusics.

Figure 5. Forest plot of effect sizes for the correlation between acoustic pitch discrimination and MBEA scale test performance.

Figure 6. Montreal database acoustic pitch discrimination plotted against MBEA scale scores. The cutoff for poor acoustic performance is 57.84 (vertical line); the cutoff for poor musical performance is 22.73 (horizontal line).