### Congenital amusia A group study of adults afflicted with a music-specific disorder

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#### Summary

The condition of congenital amusia, commonly known as tone-deafness, has been described for more than a century, but has received little empirical attention. In the present study, a research effort has been made to document in detail the behavioural manifestations of congenital amusia. A group of 11 adults, fitting stringent criteria of musical disabilities, were examined in a series of tests originally designed to assess the presence and specificity of musical disorders in brain-damaged patients. The results show that congenital amusia is related to severe deficiencies in processing pitch variCorrespondence to: Isabelle Peretz, Département de Psychologie, Université de Montréal, CP 6128, succ. Centre-ville, Montréal, Québec, Canada H3C 3J7 E-mail: Isabelle.Peretz@umontreal.ca

ations. The deficit extends to impairments in music memory and recognition as well as in singing and the ability to tap in time to music. Interestingly, the disorder appears specific to the musical domain. Congenital amusical individuals process and recognize speech, including speech prosody, common environmental sounds and human voices, as well as control subjects. Thus, the present study convincingly demonstrates the existence of congenital amusia as a new class of learning disabilities that affect musical abilities.

Keywords: congenital amusia; tone-deafness; auditory disorder; learning disabilities; music

#### Introduction

Language and music have many similarities. Notably, language and music are universal and specific to humans. Despite the complex abilities involved in both domains, linguistic and musical competence develop in the child spontaneously, without conscious effort or formal instruction. However, a few individuals suffer from severe language acquisition impairments, which are not consequent to any hearing deficiency, mental retardation or lack of environmental stimulation (e.g. Benton, 1964; Gopnik and Crago, 1991). Such a specific language impairment affects between 3 and 6% of the population (e.g. Wright et al., 1997). Considering the similarities between music and language, we can expect that a similar proportion of individuals from the general population experience music-specific impairments. Affected individuals would be born without the essential wiring elements for the development of a normally functioning system for music. This condition is variously termed tune-deafness, tone-deafness, dysmelodia or dysmusia in the literature. However, we prefer to refer to this learning disability with the less restrictive label of 'congenital amusia', because there may be as many different forms of developmental amusias as there are varieties of acquired

amusias that result from accidental brain damage in adult-hood.

Congenital amusia is a condition that has been known for more than a century, since the pioneering study published by Grant-Allen in 1878. Grant-Allen reports the case of a 30year-old man with a solid education and without neurological lesion, who suffers from a severe musical handicap. The man was unable to discriminate the pitch of two successive tones, failed to recognize familiar melodies and could not carry a tune. He exhibited an overall indifference towards music. Yet, the musical defect could not be explained by a lack of exposure to music since the man had received musical lessons during childhood (Grant-Allen, 1879). A century later, Geshwind published a similar case (Geshwind, 1984). The case was a man who came from a musically impaired family, despite their frequent exposure to recorded music at home. As a child, this man attempted piano lessons, but his teacher soon realized that he could not sing, nor discriminate between two pitches and could not keep time. Interestingly, this same subject could speak three foreign languages fluently.

Though indicative, these two studies are anecdotal since they are descriptive and not supported by systematic evaluations. Two large-scale studies were run to quantify the musical disorder. In 1948, Fry evaluated a 1200 subject sample on tests requiring the subjects to compare two notes or two musical phrases in order to detect a change in pitch. From these results, Fry estimated that 5% of the British population were amusical. This author further argued that musical memory problems as well as a difficulty in pitch discrimination might be the major determinants of congenital amusia (Fry, 1948). However, these claims were not supported by data analyses. More recently, Kalmus and Fry ran another large-scale study with 604 unselected adults who were required to detect anomalous pitches inserted in melodies (Kalmus and Fry, 1980). From these results, 4.2% of the British adult population were estimated to be amusical. However, this estimate is problematic. First, the measure lacks sensitivity since >90% of the participants were performing at ceiling. Secondly, a single measure of musical ability may have both poor validity and poor reliability. Above all, such a psychometric definition of congenital amusia is unconvincing because the sole consideration of one tail of the normal distribution on a single test does not provide convincing evidence that congenital amusia is a real affliction and not a statistical anomaly.

In summary, previous studies of congenital amusia provide valuable information regarding the nature of congenital amusia, while they do not offer a solid empirical basis. Thus, the major objective of the present study is to document the probable existence of congenital amusia by the systematic evaluation of individuals who, despite normal exposure to music and a high level of general education, failed to develop basic musical skills. These basic musical abilities rest on core mechanisms that are assumed to be shared by all members of a given society, musicians and non-musicians alike, and that allow humans to appreciate and respond to the music of their culture (Peretz, 2001). At the very least, these skills should encompass the ability to discriminate and recognize the music of the environment and, above all, to respond to it emotionally. Moreover, within limits, all humans should be able to carry a tune and to synchronize with the musical behaviour of others, by tapping along with the music and by dancing, for example. These are the basic musical abilities that will be assessed experimentally in the present study.

Based on the published findings, we expect amusical participants to show a particular deficit in discriminating musical pitch variations and in recognizing familiar melodies. As a consequence of these receptive disorders, we also expect amusical individuals to have poor singing abilities. In contrast, we have no particular predictions regarding their competence in monitoring rhythmic structure in music. Therefore, amusical subjects were tested in various abilities, tapping mostly pitch-related abilities but also rhythmic ones for comparison.

In order to assess the domain specificity of the auditory disorder exhibited by congenital amusical subjects, their ability to recognize and memorize music was compared with their ability to recognize and memorize other familiar auditory tokens, such as spoken lyrics, speakers' voices and animal cries, under identical testing conditions.

#### **Case description**

The most challenging part of the present study was to set the criteria that would allow classification of individuals as being congenitally amusical, and to find appropriate means to discover them. In what follows, we will describe (i) the procedures and the inclusion criteria used to identify amusical cases; and (ii) a summary of the behavioural assessment and self-description of the amusical participants.

#### **Recruitment** of amusical subjects

Various procedures were used, all requiring self-declaration of a handicap for music. The most effective means consisted of making announcements in the media (radio, newspapers, university local papers and vocal recording machines). However, self-declaration did not suffice. Non-musicians are prone to complain about their musical deficiencies, in general. To exclude these false alarms as well as borderline cases, we used a detailed questionnaire and focused our attention on individuals whose self-description was as close as possible to the case reports of Grant-Allen and Geshwind (Grant-Allen, 1878; Geshwind, 1984). Out of more than 100 interviews, we selected 37 individuals and tested them in the laboratory on a musical screening battery (see below). Out of these 37 potential subjects, 22 exhibited a pattern of performance that unambiguously indicated the presence of a receptive musical disorder. However, only 11 of them were willing to participate in further evaluations and were eligible because their past history fitted with the following criteria: (i) a high level of education, preferably university level, to exclude general learning disabilities or retardation; (ii) music lessons during childhood, to ensure exposure to music in a timely fashion; (iii) a history of musical failure that goes back as far as they could remember, to increase the likelihood that the disorder is inborn; and (iv) no previous neurological or psychiatric history to eliminate an obvious neuro-affective cause.

## Assessment and description of the amusical group

In order to verify the presence of a deficit in music perception and memory, all self-declared amusical subjects who met our set of stringent criteria were tested over a battery of tests designed to diagnose musical deficits in brain-damaged patients of variable age and education level. This battery contains six subtests, in which the material is conventional and kept as constant as possible across conditions. Each subtest measures the use of a musical characteristic that is known to contribute to music perception and memory. These are scale, interval and contour information on the melodic

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Table 1 Subjects' characteristics and individual scores for the amusic group

	A1	A2	A3	A4	A5	A6	A7	A8	A9	A10	A11	Controls (SD)	
												Matched	Unselected
Gender	F	М	М	F	F	F	F	F	F	F	F	16F 4M	42F 19M
Language	Fr	Fr	Fr	Fr	Fr	Е	Fr	Fr	Fr	Fr	Fr	16Fr 4A	57Fr 4A
Age (years)	41	62	57	51	71	69	58	59	53	49	57	60.2 (12.8)	45.3(17.6)
Education	19	16	19	15	17	19	15	16	19	19	17	16.5 (2.2)	13.8 (3.7)
Handedness	А	R	R	R	R	R	R	R	R	R	R	R	59R 1L 1A
I.Q.	111	116	107	100	104	112	117	108	128	110	120	_	_
M.Q.	113	135	112	114	127	130	134	114	137	114	130	_	_
Audiometry (1)													
Low frequency (250–500 Hz)	n	15–35	n	n	n	n	n	n	n	n	n		-
Middle frequency (1000-2000 Hz)	n	35–50	n	n	n	n	n	n	n	n	15–55		_
High frequency (4000–8000 Hz)	n	50-70	n	n	n	n	60–70	n	70–80	n	25-65		_
Musical battery													
Scale	76.7	60*	50*	50*	56.7*	46.7*	63.3*	53.3*	56.7*	53.3*	46.7*	90 (7.8)	91.7 (6.8)
Contour	50*	43.3*	50*	70	53.3*	46.7*	80	56.7*	66.7*	53.3*	66.7*	91.5 (6.4)	90.2 (7.0)
Interval	56.7*	56.7*	50*	50*	50*	53.3*	60*	53.3*	73.3	53.3*	73.3	88.7 (7.2)	89.3 (7.9)
Rhythm	53.3*	73.3	50*	53.3*	53.3*	76.7	76.7	63.3*	96.7	63.3*	93.3	91.7 (8.2)	91.5 (6.8)
Metric	63.3	66.7	56.7	53.3	60	76.7	60	70	73.3	73.3	70	83.5 (10.3)	81.6 (9.9)
Memory	66.7*	53.3*	50*	46.7*	40*	53.3*	73.3	50*	73.3	66.7*	80	92.8 (6.3)	89.5 (7.2)

F = female; M = male; Fr = French; E = English; A = North American; (1) = a loss expressed in dB HL for the left and right ear; n = normal; SD = standard deviation. \*Scores <3 SDs from the mean of the unselected control group.

organization dimension, and rhythm and metre on the temporal organization dimension. The sixth subtest of the battery probed memory recognition abilities. The melodic discrimination subtests as well as the rhythmic test, all require a 'same–different' classification task. The metric test requires a waltz–march classification of each musical excerpt. Finally, the memory recognition subtest appears as an incidental memory test at the end of the evaluation, by asking subjects to judge whether or not they had heard the musical selections in the previous subtests. All stimuli are computer-generated and delivered with a piano sound (for a full description see Liégeois-Chauvel *et al.*, 1998).

The musical battery has been used extensively in our laboratory and, above all, has been shown to be effective in identifying adult non-musicians with deficits in either the melodic or rhythmic dimension (Peretz, 1990; Peretz *et al.*, 1994, 1997; Liégeois-Chauvel *et al.*, 1998; Ayotte *et al.*, 2000). The conservative cut-off score of three standard deviations below the mean obtained by control subjects was used to indicate the presence of a deficit. Control subjects were 61 non-musicians (age range 14–74 years, range in years of education 7–20 years), who had no known music impairment and who were individually tested in our laboratory as neurologically intact controls for brain-damaged patients.

The performance of the 11 amusical subjects on the battery is expressed in percentages of correct responses and is presented for each individual in Table 1. When the score is below the cut-off score for a particular subtest, it is marked by an asterisk. As can be seen, all amusical subjects performed 3 SD under the mean of the 61 controls in at least two out of the six subtests. More importantly, all amusical participants failed in at least two of the three subtests involving discrimination of pitch modifications (see scale, contour and interval subtest). Performance on the rhythm subtest was more variable, with about half the subjects showing a deficit. None of the amusical participants scored below the cut-off score for the 'metric' task, probably because this task was relatively difficult for a few control subjects as well [We are currently revising the metric subtest of the battery so that all individuals (without musical impairments) achieve at least 75% correct.] Finally, eight amusical participants also suffered from a severe difficulty in memory, as indicated by their impaired performance on the incidental memory recognition test.

Thus, the presence of a musical pitch discrimination deficit is clearly supported by the results. However, it is worth mentioning that no single subtest of the battery can be used to discriminate amusical subjects from controls since, in each subtest, a few amusical subjects managed to perform in the low but normal range.

The amusical group was composed of nine women and two men, and were French speaking, with the exception of one who spoke English. Their mean age was 57 years and the mean level of education was 17 years. The higher proportion of women and of older people is probably not related to the condition of congenital amusia, but rather reflects the general characteristics of educated volunteers. One drawback of this sample is that many elderly individuals suffer from hearing problems. To assess the presence and importance of a hearing impairment, each subject underwent standard audiometric testing with a recently calibrated Beltone 9D apparatus. A summary of the outcome is presented in Table 1 along with the characteristics of each subject. When there was an impairment, the loss is expressed in dB HL for the left and right ear, respectively. As is often the case with ageing, most hearing losses are confined to the high frequency range. However, two amusical participants (A2 and A11) had a more important loss. The loss was not congenital but acquired as a result of regular exposure to loud noise during adulthood in one case (A2) and due to successive ear infections in childhood for another (A11).

However, the hearing impairment and the maturity of the amusical participants have no apparent influence on their general intellectual and memory functioning, as attested by their homogeneously high scores on standardized batteries (see Table 1). They scored above average in the WAIS-III scale (Wechsler Intelligence Scale III) and the Wechsler Memory Scale III (Wechsler, 1997*a*, *b*). Moreover, amusical participants did not report any learning disability other than for music.

According to their self-report, all amusical participants mentioned their inability to discern wrong notes in a musical passage and to sing in tune. These reports rely on peer accounts because the amusical participants are unable to perceive their own impairments. A majority (seven) reported a difficulty in recognizing musical melodies without lyrics and in dancing (eight people). Most (seven) did not appreciate music and two subjects even found music unpleasant and tried to avoid it. While all amusical participants affirmed having these difficulties as far back as they could remember, many subjects realized the scope of their problem during music classes at school. Six amusical subjects also mentioned that one of their parents (most often the mother) and certain siblings also had musical problems. However, we found members in each family who were not affected, thus discarding a familial negative attitude toward music as an explanatory factor.

### General method Matched control subjects

A group of 20 persons matched to each amusical subject in gender, age, education and musical background served as matched control participants. Sixteen control subjects were raised in the francophone culture of Quebec, while four were raised in the English culture of North America. Amusical and control participants were naive with regard to behavioural testing and were tested at the same pace. Although controls were not actively involved in music, none reported problems in the musical domain and indeed none exhibited a deficit in the screening musical battery (see Table 1). A history of alcohol abuse, psychiatric disorder or other neurological illness was grounds for exclusion. All controls were righthanded, and a hearing problem was suffered in the same proportion as the amusical participants. One control subject suffered from a hearing loss that spanned the whole frequency range, as did A2 (see Table 1), and four controls had a hearing loss in the high frequency range, as did amusical participants. All subjects' informed consent was obtained to participate in this project, which was approved by the ethical committee of the Institut universitaire de Gériatrie de Montreal.

#### Material and procedures

Most tests used to study the amusical subjects and their matched controls have been designed previously for and validated with brain-damaged non-musicians who suffered from musical impairments as a result of brain damage. Therefore, a detailed description of the tests can be found in prior published papers. The references will be provided below in the corresponding section; accordingly, only the methodological aspects that are relevant to the understanding of the testing situation will be specified here.

Stimuli were pre-recorded and delivered from a DAT Sony recorder to the subject via two loudspeakers set to a volume that was comfortable for the subject. Each subject was tested individually in our laboratory and provided his/her judgements on answer sheets. Each subject was tested in at least three sessions, each lasting ~2 h, with as many pauses as requested. The order of tests was identical for all subjects, who started with the musical screening battery described previously. They were then tested with the three types of memory recognition tests (presented in Part 2) followed by the musical pitch perception tests (presented in Part 1). The production tasks (presented in Part 3) were administered at the end.

In the following sections, the results will be presented along with the corresponding test material and procedure. Test results will be grouped in three different parts, each devoted to the behavioural assessment of a particular question regarding the functioning of amusical subjects.

### Part 1. Musical pitch perception Anomalous pitch detection task

All amusical cases reported in the literature experience a marked deficit in discerning pitch differences. This suggestion finds support in the present study since all 11 amusical participants failed in at least two out of the three screening tests that probed their ability to discriminate melodies on the pitch dimension. The results were obtained with unfamiliar melodies in a 'same-different' classification task, whereas the proportion of congenital amusical subjects in the British population was estimated with an anomalous pitch detection task and familiar melodies (Kalmus and Fry, 1980). Thus, it was deemed appropriate to test the present sample of amusical individuals with a similar test. Moreover, a variant



**Fig. 1** Mean percentage of hits minus false alarms obtained for the amusical and control group in the anomalous pitch detection task. Each dot represents an individual score.

of the anomalous pitch detection task has been used recently in a twin study and shown to tap an ability that is genetically determined (Drayna *et al.*, 2001). In order to facilitate comparisons across studies and to test further the musical pitch defect characterizing congenital amusia, we examined here the ability of our amusical group to detect a pitch anomaly in both familiar and unfamiliar melodies.

Subjects were presented with two sets of melodies. The first set comprised only familiar melodies, and the second set only unfamiliar ones. The familiar melodies were different for the French- and English-speaking participants. The 'French' version was constructed with 36 melodies (ranging in length from six to 15 notes) that are well known to Quebec French-speakers (Peretz et al., 1995); the test has been used in a prior study on acquired amusia (Peretz and Gagnon, 1999). The 'English' version consisted of 30 familiar melodies (ranging in length from seven to 14 notes) selected from a list of musical pieces that are well known to North American English-speakers (Steinke et al., 2001). The unfamiliar set corresponded to the 30 comparison melodies used in the 'scale' subtest of the musical battery. In each set, half the melodies were modified by shifting the pitch of one note by one semitone higher or lower so that the note fell out of key while it preserved the original contour. The position of the modified note varied across melodies, avoiding the first and last note positions. Each melody was presented once. After each presentation, subjects were asked to judge whether the melody contained a 'wrong note' or not.

The responses were considered as hits when the subjects responded 'yes' to a melody containing an anomalous pitch and as false alarms when responding 'yes' to an intact melody. The results are presented in Fig. 1. As can be seen, amusical subjects were performing close to chance and well below their matched controls. There is no overlap between the amusical and control distribution. The mean percentages of hits minus false alarms were submitted to an ANOVA (analysis of variance), with familiarity (familiar versus unfamiliar melodies) taken as the within-subjects factor, and groups (amusicals versus controls) as the between-subjects factor. As expected, a highly significant group effect is obtained, with F(1,28) = 162.89, P < 0.001, due to the clear deficit exhibited by amusicals compared with controls. A main effect of familiarity was also observed [F(1,28) = 35.34, P < 0.001], reflecting the higher performance observed for the familiar over the unfamiliar melodies. There was no interaction with the group factor (F < 1).

The results obtained with this anomalous pitch detection test are key for several reasons. From a practical perspective, this test clearly distinguishes amusical subjects from normal individuals, and hence may serve as a diagnostic tool in the future. The results are theoretically important because they converge with prior findings in identifying a deficiency in musical pitch perception in congenital amusia. Such a pitch defect is the most likely origin of the musical disorder, as will be argued further below and in the general discussion.

#### Sensitivity to dissonance

Another striking experience of musical pitch perception occurs when several tones sound together resulting in a harmonious blending effect. This pleasant experience is determined mainly by the ratio between the constituent frequencies. When the ratio is simple, such as between two tones lying an octave apart, the combination is considered consonant and pleasant for most listeners, including infants (e.g. Zentner and Kagan, 1996). When the ratio is complex, such as between two tones that lie a semitone apart, the resulting combination is perceived as dissonant and unpleasant by the vast majority of listeners, from an early age (for a review, see Schellenberg and Trehub, 1994). Thus, sensitivity to dissonance is a fundamental experience of music that is tightly related to the ability to perceive fine-grained pitch differences. If the latter ability is impaired, as is apparently the case in congenital amusia, then one can predict that amusical individuals will be indifferent to the presence of dissonance. The test of this particular prediction was the goal of the present investigation.

In order to assess sensitivity to dissonance, we used pleasantness judgements for a set of 24 musical excerpts that were presented in two versions (for a full description, see Peretz *et al.*, 2001). In the original version, the excerpts that comprise a melody and an accompaniment are highly consonant and are taken from pre-existing classical music (e.g. the first bars of Albinoni's *Adagio*). The dissonant version was created by shifting the pitch of all tones of the leading voice by one semitone either upward or downward. Each excerpt was presented in its consonant version and in its



Fig. 2 Mean ratings obtained in pleasantness judgements of dissonance (left panel) and in happy-sad control judgements (right panel).

new dissonant version, amounting to 48 trials. The task of the subject was to judge the pleasantness of each excerpt on a 10point scale, with 1 referring to very unpleasant and 10 to very pleasant. Since half the excerpts evoke a sense of happiness (they were all played in the major mode with a median tempo of 138) and the other half a sense of sadness (they were played in the minor mode at a median tempo of 53), these were presented in their consonant version in a separate control task. The task was to judge, on a similar 10-point scale, if each excerpt was sad (corresponding to a rating of 1) or happy (corresponding to 10). In this happy-sad distinction task, the pitch intervals (defining the mode) were not the only cue that the subject could use to recognize the emotion; the tempo was also available. To the extent that amusical subjects can derive the tempo of music, they should be able to attribute the correct emotional label to the 24 original excerpts.

The mean pleasantness ratings given by the amusical and control subjects to the consonant and dissonant version of the same excerpts are presented in Fig. 2 (left panel). As shown, control subjects judge the consonant versions as generally pleasant whereas they judge the dissonant versions as unpleasant. This distinction does not emerge in the ratings of the amusical subjects, who tend to judge all excerpts as weakly pleasant [one amusic subject (A6) did not perform this test]. The ratings were submitted to an ANOVA considering dissonance (consonant versus dissonant version) as the within-subjects factor and group as the betweensubjects factor. The analysis gives rise to a highly significant interaction between dissonance and group, with F(1,24) =52.37, P < 0.001. The interaction supports the observation that control subjects find the consonant versions much more pleasant than the dissonant versions of the same excerpts [t(15) = 12.456; P < 0.001], whereas the amusical group shows a less marked preference [t(9) = 2.123; P < 0.05]. Thus, amusical subjects appear less sensitive to dissonance than normal controls.

However, amusical subjects are able to recognize the affective tone of music to some extent. They are able to distinguish happy from sad music reliably [see Fig. 2, right panel; t(9) = 9.692; P < 0.001], although their judgements are less extreme than those of their controls. This difference is supported by a significant interaction between structure (major/fast excerpts versus minor/slow excerpts) and group, with F(1,23) = 5.78, P < 0.025.

Altogether, the results obtained with emotional judgements are consistent with those obtained in non-emotional tasks. Amusical subjects are unable to perceive and interpret musical pitch differences normally. Yet, this is not the result of poor auditory attention or of a deficient affective system in general, since they are able to infer the happy or sad tone of music in a rather consistent manner.

## Discrimination of pitch variations (intonation) in speech

Fine-grained discrimination of pitch is probably more relevant to music processing than to any other domain. Speech intonation contours, for example, use variations in pitch that are larger than half an octave, to convey relevant information. In contrast, melodies use mostly small pitch intervals (of the order of a 1/12th or 1/6th of an octave, which correspond to the semitone and whole tone, respectively). Therefore, a degraded pitch perception system may compromise music perception, but leave speech prosody unaffected.

To assess this hypothesis regarding the domain specificity of the pitch defect experienced by amusical subjects, we again exploited experimental tests that have been used previously with brain-damaged amusical patients (Patel *et al.*, 1998). These tests are constructed by computer-editing two basic sets of sentences so that they differ from each other



Fig. 3 Mean percentage of hits minus false alarms and standard errors obtained, in each group, for spoken sentences (left panel) and non-speech derivations (right panel) as a function of the location of the pitch discrimination change.

only by local pitch changes. In the first set, the change affects the last word by marking a rise in pitch so as to indicate a question (e.g. He speaks French?) or a falling pitch (e.g. He speaks French) to indicate a statement. The pitch change is in the order of six to seven semitones (range three to 11) for questions in French and English, respectively. In the statements, the pitch fall is of two to three semitones. In the second set of sentences, the pitch difference (of an average magnitude of eight semitones) affects an internal word of the sentence to mark emphatic stress such as in 'SING now please!' (upper case letters indicate the stressed word) and 'Sing NOW please!' Both sets were naturally spoken by a native female speaker and exist in French and English; each set comprises 15 or 16 pairs of sentences, depending on language.

The sentences were first presented in isolation, and subjects judged whether the sentence indicates a statement or a question for the first set of sentences differing by their final pitch rise, or they indicated which word bore the stress for the second set of sentences. These two tasks were relatively easy to complete for both amusical and control subjects, who scored 94 and 98% correct, respectively, in the final pitch change condition, and 87 and 90%, respectively, in the internal pitch change condition.

The same sets of sentences were also presented in pairs in a 'same-different' classification task. For example, when subjects were presented with the pair 'He speaks French?' followed by 'He speaks French', they were expected to respond 'different'. The only cue available for the discrimination was the presence or absence of a pitch change on the final word in 32 trials (30 in English) and in the location of the internal pitch change in 32 additional trials (30 in English). An equal proportion of trials contained no change. No feedback was provided to the subject. Again, amusical subjects performed slightly less well than the control subjects (see left panel in Fig. 3) but not significantly so [F(1,28) = 1.41]. Thus, amusical subjects do not seem to be impaired in processing speech intonation, even when these prosodic variations are limited to pitch changes as studied here.

When all linguistic information is removed from the sentences by a process of computer analysis and synthesis (for details of the editing of these non-speech analogues, see Patel et al., 1998), a different picture emerges. With the nonspeech derivations of the sentences, amusical subjects show evidence of a deficit compared with normal controls, as can be seen in the right panel of Fig. 3. Yet, the only difference between the speech results (left panel) and the non-speech analogues (right panel) lies in the acoustic waveform of the stimuli, not the size of the pitch differences. This difference significantly affected the performance of amusical subjects, as confirmed by an overall ANOVA taking material (speech versus non-speech) and type of pitch change (final versus internal pitch change) as the within-subjects factors, and groups as the between-subjects factor. The interaction between all three factors reached significance [F(1,28) = 5.04, P < 0.033]. This was due to the poorer performance of the amusical subjects in the non-speech condition relative to normal controls [F(1,28) = 12.88;P < 0.001]. Non-parametric tests (Mann–Whitney U tests) yield similar results.



Fig. 4 Mean percentage of correct responses and standard errors obtained for each group in the identification of melodies, lyrics, speakers' voices and environmental sounds. Naming and recognition scores are represented separately.

One can conclude from this set of tests that congenital amusia does not compromise interpretation and discrimination of speech intonation. However, this spared area of performance is contingent upon the presence of linguistic information. When linguistic cues are removed from the signal, amusical subjects exhibit reduced performance. The latter finding suggests that the pitch defect of amusical subjects is not limited to music but can extend to other auditory patterns varying on the pitch dimension, provided that amusical subjects cannot use speech cues to support discrimination.

#### Part 2. Specificity of the musical disorder

The tests using intonation patterns in speech suggest that amusical subjects may have difficulties in the processing of auditory patterns other than musical ones. Yet, following the literature as well as self-reports of amusical subjects, the disorder seems limited to the musical domain. In order to delineate the auditory domains in which amusical subjects seem to be at a disadvantage compared with normals, they were tested here with auditory meaningful stimuli pertaining to speech, human voices and environmental sounds. Care was taken to make cross-domain comparisons in identical experimental conditions so as to minimize the contribution of other factors to the tasks. In doing so, we relied heavily on prior work that succeeded in demonstrating the domain specificity of the musical impairments observed in brain-damaged patients (see, in particular, Peretz et al., 1994, 1997; Peretz, 1996; Ayotte et al., 2000).

#### Naming and recognition of tunes, lyrics, voices and environmental sounds

The identification of a familiar auditory pattern by name is one of the most demanding tasks, particularly for non-verbal material. Yet, it is one of the rare tasks that allows the rapid assessment of the integrity of a processing system from the analysis of the acoustic input up to the name retrieval via appropriate contacts in memory. This is why identification tasks are used regularly in neuropsychological settings. In addition, this explains why we administered this test first so as to assess the domain specificity of the difficulties experienced by congenital amusical subjects.

Subjects were presented with auditory stimuli from four different domains, but blocked by domain. The musical block consisted of 52 melodies (without lyrics) from familiar folk songs (Peretz et al., 1995; Steinke et al., 2001) and presented one at a time. After each presentation, the subjects were requested to name the tune. In the case of failure, they were presented with four written titles from which to choose; the foils were of the same genre (e.g. all titles were Christmas songs). The lyrics block comprised 25 spoken lyrics from the same pool of well-known songs. However, care was taken to select lyrics in which content words could not cue the title (e.g. 'prête-moi ta plume pour écrire un mot' for the song name 'Au clair de la lune' or 'for nobody else gave me a thrill' of the song name 'It had to be you'). In the case of failure, they were presented with four written titles semantically related to the lyrics excerpt. The voice block involved 33 speech excerpts pronounced by famous public figures for Quebec residents. The stimuli were edited so as to remove

any context word that could cue the speaker's professional activities. Three choices of names of the same sex and age were presented in the case of a naming failure. Finally, 45 environmental sounds, including animal cries, transportation noises, human noises and indoor noises, were presented in a block. In case of a naming failure, subjects could choose between four pictures of the same category (e.g. all pictures would be means of transportation).

The mean percentages of correct naming scores and global scores, which add the correct naming to the correct name choices, are presented in Fig. 4, for each domain. The naming and global scores are highly similar. By both scoring procedures, amusical subjects were disproportionately impaired in music identification relative to the other domains and to control performance. With the exception of one outlier, all amusical subjects were able to retrieve the name of the song corresponding to the spoken lyrics. This finding is important because it shows that amusical subjects have learned the songs although the musical part is problematic for them.

The outcome of the ANOVAs performed separately on the naming and global scores confirms these observations. Each ANOVA was computed with material (tunes, lyrics, voices and environmental sounds) as within-subjects factor and group (amusics and controls) as the between-subjects factor. An interaction was obtained between material and group [*F*(3,69) = 17.26 and 12.23, *P* < 0.001, on global and naming scores, respectively]. The interaction was due to the fact that the amusical group only performed below the control group in the music identification test [t(29) = 8.526 and 6.609, P < 0.001, for the naming and global scores, respectively]. No difference was observed between groups for the other materials [with t(29) = 0.551 and 1.494 for lyrics, n.s.; t(24) = 0.019 and 0.982 for human voices, n.s.; and t(23) = 0.203 and 0.073 for environmental sounds, n.s., for the naming and global scores, respectively]. The nonparametric statistical analyses (Mann-Whitney U test) yielded the same results.

The results confirm the self-report of amusical subjects in showing a selective problem in recognizing the melodic part of songs. Otherwise, the amusical subjects appear to recognize lyrics, a speaker's voice and environmental sounds as easily as everybody else. The disorder appears to be musicspecific.

# Separate recognition of tunes and lyrics from songs

An important result in the previous identification tests was to find a clear dissociation in all but one amusical subject between the recognition of lyrics, which was intact, and the recognition of tunes, which was impaired, although both components were learned together in songs. To assess this particular dissociation further in a less demanding task, we used here a binary familiarity decision. Requiring a feeling of knowing without identification may reveal residual recognition abilities. To this aim, a familiarity decision task was devised for tunes and lyrics, separately. Half the stimuli came from familiar songs and half were unfamiliar. The unfamiliar melodies were taken from the same repertoire of folk songs but are unknown to the subjects because they are no longer sung (for details about stimuli selection, see Peretz *et al.*, 1998). The task is simply to indicate whether or not each song part sounds familiar.

The responses were considered as hits when the subjects responded 'familiar' to familiar excerpts and as false alarms when responding 'familiar' to unfamiliar excerpts. The mean percentages of hits minus false alarms obtained by each group in the two familiarity decision tests are presented in Fig. 5 (the negative values indicate a greater number of false alarms than of hits). The data were submitted to an ANOVA with material (melodies versus lyrics) as the within-subjects factors and groups as the between-subjects factor [one amusical subject (A6) did not perform this task]. An interaction between group and test was obtained [F(1,24) = 11.05, P < 0.003]. The amusical group was again only impaired in the melody condition relative to the control group [t(24) = 4.603; P < 0.001]. Amusical subjects did not perform significantly below normals in the lyric condition (n.s.), despite the presence of a very poor score obtained by the same outlier as previously. The same results were obtained by way of Mann–Whitney U tests.

### Memory recognition for tunes, lyrics and environmental sounds

One important question regarding the failure of amusical subjects to recognize familiar melodies is to what extent they are able to relearn these melodies in the laboratory. The results obtained in the screening battery indicate an overall deficit in memorizing novel music (see Table 1). However, in this memory recognition test, memorizing melodies was incidental. Incidental encoding of music may not reflect the optimal performance of amusical subjects since they were not fully engaged in memorizing the material. Thus, it remains possible that amusical subjects are able to memorize melodies if they are explicitly told to do so. We also need to assess whether memorizing familiar music is possible. Although the amusical group has difficulties in recognizing music as familiar, they might remember it nonetheless. For example, when presented with the music of 'La vie en rose', they might recognize it as music which they had heard in the laboratory, without recognizing it as familiar. Evaluation of these possibilities was the goal of the following experiment.

To test memory, we used the same recognition memory test with three different materials as previously validated with brain-damaged patients (Peretz, 1996; Peretz *et al.*, 1997). In the presentation phase, 20 auditory targets are presented one after the other. The subject's task is to memorize each of them. After a short pause, the test phase occurs, during which the 20 targets are mixed randomly with 20 lures; the subjects' task is to tell for each stimulus whether or not they have heard it in the study set. No feedback is provided to the subject. In one form of the test, all stimuli consist of familiar tunes. Subjects performed the same task for two non-musical materials, one including lyrics and one involving environmental sounds, similar to those used in the previous tests of identification. The three memory recognition tests were performed in different testing sessions.

The responses were considered as hits when the subjects responded 'yes' to studied stimuli, and as false alarms when responding 'yes' to non-studied stimuli. The mean percentages of hits minus false alarms obtained by each group in the three memory recognition tests are presented in Fig. 6. These scores were submitted to an ANOVA with material (melodies, lyrics and environmental sounds) taken as the within-subjects factor and group as the between-subjects factor. As shown in Fig. 6, the amusical group scored significantly below the control group on the tune recognition test [t(29) = 5.994; P < 0.001] but not so on lyrics [t(29) = 0.502; n.s.] and environmental sounds [t(29) = 1.541; n.s.]. The selectivity of the impairment to the musical domain was supported by the presence of an interaction between material and group [F(2,58) = 30.86, P < 0.001].

To conclude, subjects with congenital amusia perform poorly in all tasks requiring recognition and memory of melodies. The musical deficit is apparent in all tasks, from the most difficult situation that requires naming a well-known tune, to the least demanding task requiring a familiarity decision. Even re-learning is unsuccessful, when provided with a single study episode (i.e. in the last condition referred to as the memory recognition condition). The performance of the amusical subjects is low, but variable; yet, it is remarkably consistent across the three recognition tasks involving familiar melodies. Pearson correlations (with n = 10) are 0.90 between the scores obtained in identification (global score) and familiarity; 0.77 between familiarity and memory recognition; and, finally, 0.79 between identification and memory recognition (all P < 0.05). In contrast, under identical testing conditions, the same amusical subjects show no particular difficulty in identifying familiar songs on the basis of their spoken lyrics, or in recognizing and memorizing non-musical auditory events such as common environmental sounds and speakers' voices. Clearly, the amusical participants do not suffer from general difficulties in memory or attention in the auditory domain. They suffer from a highly selective memory deficit that seems limited to music.

#### Part 3. Musical production tasks

Our sample of amusical participants were selected because they report and show evidence of severe perceptual impairments for music. Yet, congenital amusical subjects are notoriously famous for singing out of tune. They are usually detected on this basis. In the case of our sample, the production deficit can be expected to arise as a consequence

Amusicals



Fig. 5 Mean percentages of hits minus false alarms obtained, for each group, in familiarity decisions for melodies and spoken lyrics of songs. Dots represent individual results.

of their poor perceptual and memory system. To assess this rather obvious prediction in a controlled manner, each amusical subject (as well as their controls) was encouraged to sing three songs into a microphone. To assess their potential to synchronize with music (for dancing, for example), they were also assessed in their ability to tap out the beat of three different pieces of music while being videotaped.

The ability to carry the tune of 'Au clair de la lune', 'Frère Jacques' and 'Vive le vent', which are highly familiar children's songs in French-speaking cultures, was assessed in two conditions. In the first condition, the lyrics were provided in print and the participant had to sing the corresponding tune from memory. Since amusical subjects show poor memory for those tunes, as documented previously, their ability to repeat these same tunes, after the experimenter, was checked in a second condition. The singing production of only seven amusical participants could be recorded (A3 and A6 could not participate and A9 and A11 refused to sing in the repetition condition). Controls were tested in equal number in the same conditions. All amusical and control renditions were recorded on audiocassette and then mixed randomly. There were two cassettes: one containing singing from memory and the other sung repetition.

The ability to synchronize with music was assessed with the first 30 bars of pre-recorded music. The recordings were selected in three different genres; one piece was classical (Ravel's Bolero; duration 1 min 22 s), another was disco ('Stayin'alive' from the Bee Gees; duration 1 min 28 s) and the third was folk ('reel des soucoupes volantes' from the Bottine Souriante; duration 1 min 11 s). Subjects were required to tap in time with each piece of music, and to do so as regularly as possible with their dominant hand (so as to avoid complicated or syncopated rhythms). All amusical and control tapping hands were filmed and were then copied in a random order onto another cassette.

The three tapes (two audio- and one videotape) were judged by six judges (four musicians and two non-musicians) who were blind to the classification of the participants. The judges evaluated each performance on a 10-point scale, where 1 meant 'very poor' and 10 meant 'very good'. They also had to decide if each recording was produced by an amusical subject or not. The ratings were averaged for the three songs produced by the same subject in the same condition. The agreement between judges was high, since correlations between all pairs of judges were significant in each condition (each r being >0.76; P < 0.01). The results are presented in Fig. 7. They were assessed by non-parametric Mann-Whitney U tests that confirm the presence of a clear distinction between amusical and control performance in each production condition (U = 82, 63 and 80 for singing from memory, repetition and tapping, respectively). Across conditions, amusical renditions were judged to be rather poor compared with controls. Moreover, the judges were generally accurate in their classification of each production as being from an amusical or non-amusical person. Out of the 74 amusical productions, 42 were correctly attributed to an amusical performer by at least four judges, whereas none of the 87 control productions were judged as such (the difference is significant with  $\chi^2 = 63.89$ , P < 0.001). However, the classification is not perfect since one amusical individual (A9) managed to produce an acceptable performance in each condition, by both scoring procedures.

In order to evaluate which musical aspect of the vocal production was most affected in amusical singing, we asked two further musicians to judge blindly the accuracy of the



Fig. 6 Mean percentages of hits minus false alarms obtained, for each group, in the memory recognition test as a function of the material presented. Dots represent individual results.

rendition in terms of the pitch variations and the temporal variations, separately. The judges provided their ratings on different 10-point scales (with 1 meaning very poor and 10 very good). The ratings were averaged for the three songs in each singing condition and assessed for consistency by Pearson correlations. The ratings were highly correlated, with r = 0.94 and 0.92 for the pitch and time dimension, respectively (P < 0.01). Amusical singing was judged to be more impaired on the pitch dimension (with a mean rating of 3.9 across conditions) than on the temporal dimension [mean rating = 6.4; t(8) = 6.726, P < 0.001]. However, a similar trend also emerged for the control performance, with a better rendition of melodic than rhythmic aspects [with 8.9 and 8.2 mean ratings, respectively, t(9) = 2.289, P < 0.05].

As expected, amusical singing and tapping performance is impaired as compared with normal performance. The difficulties were judged to affect mostly the accuracy of pitch variations. However, the problem was not limited to the pitch dimension since the rhythmic aspect of amusical subjects' singing was not highly rated and, above all, their tapping performance was generally not well synchronized with the music. Finally, and more surprisingly, one amusical subject was judged to perform normally in these tasks. This spared performance may be genuine, pointing to a non-trivial dissociation between perception and performance. Alternatively, this exceptional performance may simply reflect a lack of sensitivity of the crude measures of performance considered in the present study.

#### **General discussion**

This study suggests that congenital amusia is not a myth but a genuine learning disability for music. The systematic evaluation of 11 adults, who reported themselves to be severely handicapped in the musical domain despite their efforts to learn it, largely confirms the presence of an underdeveloped system for processing music. Amusical individuals are severely impaired in music discrimination and recognition tasks. These impairments cannot be explained by hearing losses, since they all have, or grew up with, normal audiometry. The musical disorder cannot be explained by a lack of exposure since all amusical participants had music lessons during childhood and were raised in families in which a few siblings are musically normal. Finally, the musical deficit cannot be ascribed to some general cognitive slowing since all amusical participants have reached a high level of education. The musical disorder appears as an accidental disturbance in an otherwise fully normal cognitive and affective system.

One remarkable characteristic of the amusical condition is the selectivity of the disorder. The deficit appears highly specific to the musical domain. Amusical subjects retain the ability to process non-musical material as well as their matched controls. In the present study, amusical subjects were shown to interpret intonation in speech properly, to identify well-known figures from their voice alone and to identify and recognize common environmental sounds, such as animal cries and ringing sounds. With the exception of a single amusical subject, they all identified and recognized familiar songs when hearing the first lyrics. This high level of achievement in the auditory domain stands in sharp contrast to the rather poor level of functioning displayed by amusical individuals in recognizing and memorizing musical patterns. The disorder appears to be music-specific.

Having established congenital amusia as a real pathological condition, we should now turn to plausible accounts of the observed deficiency. Presently, we can only offer functional explanations, derived from the present behavioural



Fig. 7 Average ratings and standard errors obtained by each group in singing from memory, sung repetition and in keeping time with the music.

studies. Although we construe congenital amusia as resulting from a slight disruption in the wiring of the auditory cortex, we presently are unable to support this claim because the search for these neural anomalies will require sophisticated brain imaging studies. In this endeavour, specifying the functional origin of congenital amusia is essential because it may narrow down the possible neural loci to a sizeable set of circuitries that can be inspected further for the presence of an anomaly.

On the basis of the present behavioural results and in line with the literature, we propose that one likely origin for congenital amusia is related to a deficiency in musical pitch recognition. Indeed, all amusical participants score below the normal range in the discrimination of musical stimuli that differ on the pitch dimension, while a majority of them succeed in discriminating the same stimuli when these differ in temporal structure. This difficulty in detecting pitch-related changes extends to dissonance, for which amusical subjects show little sensitivity. The pitch-related defect also extends to the detection of an anomalous pitch inserted in an otherwise conventional melody. This task is particularly sensitive to the presence of amusia since there was no overlap between the normal variations and the scores of the amusical subjects. The test is diagnostic in the sense that it provides a behavioural marker of congenital amusia. Interestingly, the detection of an anomalous pitch in conventional melodies is a test that is very similar to that originally used by Kalmus and Fry to discover congenital amusical subjects in the general British population (Kalmus and Fry, 1980). More importantly, the test has been shown to tap an ability that is genetically determined in the general population (Drayna et al., 2001). Therefore, it is tempting to propose that heritability of pitch recognition abilities can also be demonstrated by its deficiency, and that its manifestation is congenital amusia.

It is important to note that the pitch defect of amusical individuals does not seem to compromise music exclusively. The impairment extends to the discrimination of intonation patterns, when all linguistic cues are removed. This observation suggests that the pitch deficiency experienced by amusical individuals is not music-specific but is musicrelevant. In effect, as mentioned previously, fine-grained discrimination of pitch is probably more relevant to music than to any other domain, including speech intonation. Music is probably the only domain in which fine-grained pitch discrimination is required for its appreciation. Accordingly, a degraded pitch perception system may compromise music perception but leave other domains, such as speech intonation in which meaningful pitch variations are coarse, relatively unaffected. Yet, the same pitch-tracking mechanism may subserve both domains. The validity of this prediction currently is under closer examination in our laboratory, where we are studying single amusical cases in detail. The current evidence is largely consistent with the notion that pitch is essential to music. More specifically, a drastic pitch perception defect has been documented with psychophysical methods in one amusical participant (A1; Peretz et al. 2002).

Similarly, a deficit in monitoring pitch changes, but not timing changes, in speech derivations has been studied further and isolated in another case (A6; Hyde *et al.*, 2001).

However, the fine-grained pitch discrimination disorder is not the only impaired musical ability in congenital amusia. As mentioned above, amusical subjects have memory problems for music, some are even impaired in discriminating melodies by their rhythm, and most have difficulties in keeping time to a musical beat. All these tasks require an accurate representation of musical time, not solely musical pitch. There are several possible explanations for the presence of this myriad of musical deficits. The explanation that we presently favour, but for which we do not yet have empirical support, is that the ensemble of musical deficits are cascade effects of a faulty pitch processing system, i.e. fine-grained pitch perception might be an essential component around which the musical system develops in a normal brain.

Taken together, the neuropsychological evaluation of selfdeclared amusical adults has provided a framework for the diagnosis of congenital amusia and has served to delineate the nature and specificity of the disorder. Although there have been limited explorations into developmental musical disorders in the past, the research enterprise constitutes a rich study avenue. At the very least, continued effort in understanding the causes of congenital amusia should shed light on the question as to whether or not music processing corresponds to a genuine specialization of the brain. From an educational perspective, knowledge of every aspect of congenital amusia should enrich the current view of other forms of learning disabilities such as dysphasia and dyslexia. The broader the context in which learning disabilities are viewed, the more likely we are to understand their causes.

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