Congenital Amusia: A Disorder of Fine-Grained Pitch Discrimination

Case Study

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Summary

We report the first documented case of congenital amusia. This disorder refers to a musical disability that cannot be explained by prior brain lesion, hearing loss, cognitive deficits, socioaffective disturbance, or lack of environmental stimulation. This musical impairment is diagnosed in a middle-aged woman, hereafter referred to as Monica, who lacks most basic musical abilities, including melodic discrimination and recognition, despite normal audiometry and above-average intellectual, memory, and language skills. The results of psychophysical tests show that Monica has severe difficulties with detecting pitch changes. The data suggest that music-processing difficulties may result from problems in fine-grained discrimination of pitch, much in the same way as many language-processing difficulties arise from deficiencies in auditory temporal resolution.

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Introduction

Music, like language, is a universal and specific trait to humans. Similarly, music appreciation, like language comprehension, appears to be the product of a dedicated brain organization (Peretz and Morais, 1993). Accordingly, as with language, normal acquisition of musical competence is expected to recruit and fine tune distinct neural networks in the human brain. In the advent of a slight but congenital neural deviation, selective deficits of learning may occur. Indeed, some individuals suffer from language-specific disorders, and a large research effort has been undertaken to understand the origin and varieties of these disorders. Some researchers claim that language impairments arise from failures specific to language or cognitive processing (Studdert-Kennedy and Mody, 1995). Others hold that language deficiencies result from a more elemental problem that makes individuals unable to hear fine acoustic temporal changes (Tallal and Piercy, 1973). Nothing comparable has been initiated in the musical domain.

The possibility that certain persons are born with a specific musical deficit has been envisaged for more than a century (Grant-Allen, 1878; Geshwind, 1984). However, the evidence rests on anecdotal case descriptions. In this paper, we present the study of a female volunteer, Monica, who manifests a music-specific disorder, sometimes called tone deafness but for which we prefer the term "congenital amusia". The term better reflects the likelihood that there are multiple forms of music developmental disorders, as there are various patterns of acquired amusia resulting from brain accident (Marin and Perry, 1999).

Case History

Monica declared herself as "musically impaired" in response to a newspaper advertisement. She was selected as the most clear-cut case among 37 volunteers with similar problems who were tested in our laboratory. Monica is a French-speaking woman in her early forties, currently completing a Master's degree in Mental Health after having practiced as a nurse for many years. She has no psychiatric or neurological history or any hearing loss. A thorough and detailed audiological examination failed to reveal any anomaly in the ear. The audiometry, tympanometry, and otoemissions yielded normal results.

A quantitative volumetric examination of Monica's brain with magnetic resonance imaging was carried out after transformation by linear scaling and rotation into the standardized stereotaxic space of Talairach. Inspection of the scan did not reveal any remarkable anatomical abnormalities. There was no indication of subtle changes or atrophy within the region of the auditory cortex. The volume of both the left (4028 mm³) and right (1298 mm³) Heschl's gyrus lies within 2 SD of those (2363 ± 963 and 1626 ± 534 mm³) of 40 normal unse-
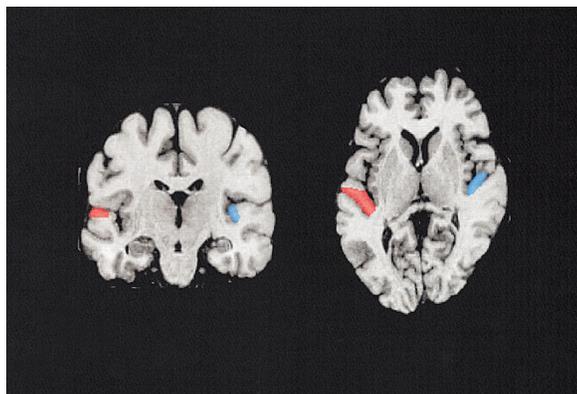
Figure 1. Magnetic Resonance Images of Monica's Brain

The left and right images show coronal and horizontal sections, respectively, through the temporal lobes. The MRI has been linearly transformed and rotated into the standardized stereotaxic space of Talairach. Heschl's gyri are labeled in color.
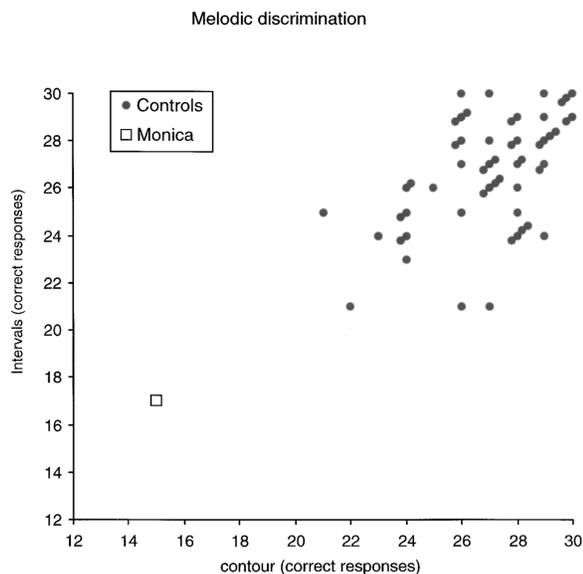

Figure 2. Melodic Discrimination

Monica's scores in the contour and interval subtests of the musical battery relative to the individual scores of 57 nonmusicians (age range: 14–74 years. Range in years of education: 7–20 years), who have no known impairment for music. The chance level is 15.

Tests and Results

To confirm Monica's disorder and to determine whether the deficit is specific to music, we evaluated her with numerous tests that were initially designed for the as-

lected subjects (Penhune et al., 1996; see Figure 1). Similarly, the volumes of secondary auditory cortices found within the planum temporale in the two hemispheres (3410 and 4916 mm^3 in left and right, respectively) were within 1 SD (4199 ± 1366 and 4105 ± 1562 mm^3) of those of a normal population (Westbury et al., 1999). These findings do not exclude the possibility that abnormalities exist at a microstructural level, but they do indicate that there is no obvious cortical atrophy or pathology.

Monica scores above average in a standard intelligence test: her WAIS-R Intellectual Quotient is 111. She has an excellent memory for both verbal and nonverbal information: her Wechsler Memory Quotient is 113. Her working memory is also normal, as measured by the digit span test (with a forward span of 7 and a backward span of 4). This span score corresponds to the 53rd and 81st percentile in the intelligence and memory scale, respectively. Nonetheless, Monica suffers from a lifelong inability to recognize or perceive music, sing, or dance. This musical handicap has persisted despite the fact that Monica was given the opportunity to develop at least some musical competence during childhood and adolescence. She was involved in a church choir as a child and was later involved in a high school band. On both occasions, Monica did these musical activities under social pressure. Monica now reports that she does not like to listen to music because it sounds to her like noise and induces stress. She admits, however, that she came to realize her shortcoming more acutely after her recent marriage to a college music teacher. It is possible that her musical disability might have a genetic origin, because other family members (her mother and brother, but not her father and sister) are reported to be similarly impaired, although they have not been formally tested.

sessment of brain-damaged patients with probable acquired amusia. Monica was then tested for the presence of a low-level pitch perception deficiency. This seems quite likely since fine-grained discrimination of pitch is probably more relevant to music than to any other domain, including speech intonation. Speech intonation contours, for example, use variations in pitch that are larger than half an octave to convey relevant information (Patel et al., 1998). In contrast, melodies mostly use small pitch intervals, of the order of 1/12 or 1/6 of an octave (Vos and Troost, 1989). Therefore, a degraded pitch perception system may compromise music perception but leave speech prosody unaffected.

Is Monica Amusic?

Monica was evaluated with a battery of tests that measure the use of musical characteristics that are known to contribute to the perception and memory of unfamiliar but conventional music. In the battery, the same pool of novel monodies are arranged in multiple tests, of which two deal with pitch variation discrimination and two with temporal variation discrimination. The tests have been used extensively and have been shown to be effective in identifying adult nonmusicians with deficits in basic musical abilities (Ayotte et al., 2000; Liégeois-Chauvel et al., 1998; Peretz, 1990). Monica's performance on most tests of the battery lies at chance level, well below the traditional cutoff score of 3 SD below the mean obtained by control subjects.

Monica's performance relative to the normal distribution is illustrated in Figure 2, with the data obtained in two essential subtests that explore the processing of sequential pitch variations, the focus of the present study. In these two subtests, two types of manipulation were applied to the same tone in the short monodies. One ma-

nipulation consisted of creating a *contour*-violated alternate melody by modifying the pitch of one note so that it changed the pitch direction of the surrounding intervals while maintaining the original key. The other manipulation consisted of creating a contour-preserved, but *interval*-violated, alternate melody by modifying the same critical pitch to the same extent (in terms of semitone distance) while maintaining the original contour and key. These melodies were used in the contour and interval subtests, respectively. Each subtest consists of 15 pairs made of identical melodies and 15 pairs of different melodies. Only the nature of the pitch change inserted in the melodies distinguishes the contour and interval subtest. Subjects are required to judge, for each pair, whether the target and the comparison sequence are the same or different. As can be seen in Figure 2, Monica's scores indicate her inability to discriminate between melodies on the basis of both their global contour and local interval pattern.

Monica's musical deficit extends to the discrimination of rhythm. In the rhythmic test, the same basic set of monodies as those used in the contour and interval subtests serve for discrimination. Only the nature of the change inserted in the comparison melody differs by involving a rhythmic modification. The manipulation consists of changing the duration values of two adjacent tones so as to change rhythmic grouping by temporal proximity while keeping the meter and the total number of sounds identical. This can be done either by changing two quarter notes for a dotted quarter and an eighth note, or by interchanging the order of two successive but different duration values (e.g., a half note followed by a quarter note becomes a quarter note followed by a half note). Thus, the only cue available for discrimination is the rhythmic pattern. The task also requires a "same-different" classification of 30 melody pairs.

On this rhythm discrimination subtest, Monica scores at chance level (with 16 correct responses out of 30, while the controls' mean is 27.4 with a SD of 2). However, she is able to derive the metrical pattern of the musical sequences to some extent since she scores in the low but normal range when classifying the same melodies as a march or a waltz (with 19 correct responses out of 30; controls' performance range: 18–30). The monodies were the two-phrase versions of the one-phrase sequences used in the other tests. The 30 melodies differed in meter, since half of these were written in a duple meter and half were written in a triple meter. This difference in meter induces the perception of a periodic alternation between strong and weak beats every two or three beats (as in a waltz: ONE, two, three, ONE, two, three,...) depending on its meter. No accent (i.e., intensity cue) is present in the music itself.

Monica's difficulties with rhythm were, however, not further examined. Her deficiency in processing melodic variations was studied in more detail. The first step consisted of documenting the perceptual basis of her problem in the discrimination of sequential pitch variations. To this aim, Monica was tested with a pitch anomaly detection task. She was presented with the same melodies as those used in the musical battery, but, this time, she was asked to detect the presence of a "wrong note" (that is, a note that was deliberately played out of key in half the melodies) by giving a "yes" or "no" response.

Monica's performance was at chance (17 out of 30 correct responses); again, far below that of unselected controls (mean = 25.1; SD = 2.5). Thus, Monica seems to suffer from a perceptual defect and not from a general deficiency in working memory for musical material.

Monica's perceptual disorder may account for her poor recognition of highly familiar music. Presented with a written forced choice among four possible titles, she is able to recognize only 22 out of 52 melodies that are familiar to most people in Quebec (Peretz et al, 1995). As expected, the result is much inferior to the scores obtained by two control groups. The first group consisted of four women of Monica's age and education level but without music-related symptoms, referred to as "matched controls" (mean = 51; $\chi^2 = 36$, p < 0.001), and the second group consisted of 29 unselected control adults (mean = 50; $\chi^2 = 32$, p < 0.001). Thus, Monica failed to learn the songs and melodies that everyone else can easily identify. This recognition failure cannot be explained by inattentiveness or a poor auditory system in general, since Monica is able to identify without hesitation 30 of 33 voices of well-known speakers, as do matched controls (mean = 30.3; SD = 3.1). The speakers can only be identified from voice characteristics, due to the fact that the speech excerpts were selected so as to contain casual speech without any contextual word cues.

Is the Disorder Specific to the Musical Domain?
Monica's deficit seems limited to music, as suggested by her intact ability to recognize voices. In order to further assess the domain specificity of Monica's impairment in comparable testing conditions, we asked her to learn 20 melodies taken from familiar songs, presented auditorily, one at a time. In a subsequent recognition test phase, Monica was asked to distinguish the studied targets from 20 unstudied (but equally familiar) melodies with which they were mixed. For comparison, Monica was also asked to learn and recognize 20 spoken lyrics (taken from the same familiar songs) and 20 environmental sounds (e.g., a barking dog). The three tests were performed in different sessions (as in Peretz, 1996). Monica performed at chance (with 52% of correct responses) for melodies and much worse than controls (with 84% and 82%, for matched and unmatched controls, respectively; $\chi^2 = 22$ and 19, p < 0.001). In contrast, her scores are comparable to those of controls for the nonmusical materials, whether song lyrics (85% correct; controls' mean = 86; SD = 8) or nonverbal environmental sounds (88% correct; controls' mean = 90; SD = 6). These results illustrate the domain specificity of Monica's musical impairment.

**Is the Musical Impairment Due
to Poor Pitch Discrimination?**
Monica's impaired monitoring of pitch errors in melodies led us to assess the presence of a low-level deficiency in pitch perception. To assess the presence or absence of a pitch defect, we trained Monica to respond to a pitch change inserted in a five-tone sequence. Monica was asked to respond "yes" whenever she detected a pitch change on the fourth tone in an otherwise constant pitch tone sequence and to respond with a "no" when unable to detect a pitch change. Feedback was provided
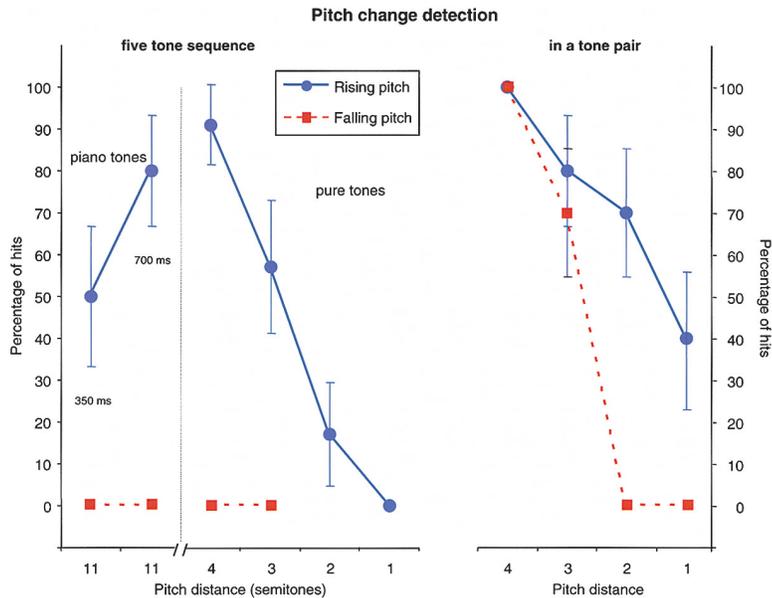
Figure 3. Pitch Change Detection

Monica's percentages of correct detection of a pitch change either on the fourth tone in a five-tone sequence (left panel) or on the second tone of a pair (right panel) as well as the corresponding standard error, for each pitch distance. False alarms are not reported, because Monica mistakenly detected a change in an identical tone sequence only once.

after each trial. The results are reported in Figure 3 (left panel).

As can be seen, Monica can detect a pitch change of 11 semitones if and only if the pitch change is rising, not when it is falling. The pattern is similar whether pure tones or piano tones are used and whether tone duration is longer (700 ms) or shorter (350 ms). With pure 700-ms tones, the only tones to be judged "intelligible" by Monica, she can barely detect a rising pitch change as large as two semitones. She is at chance for one-semitone changes. Her performance is slightly better when presented with pure-tone pairs (see the right panel in Figure 3). Yet, she is still at chance for descending pitches of two semitones.

The difference between the tone sequence and the tone pair condition cannot be explained by a working memory problem. Pitch changes are identical in the tone sequence and the tone pair; the only difference lies in the repetition of a constant pitch in the sequence context. This tone repetition would be expected to facilitate pitch change detection by providing more reference tones and twice as many cues in terms of pitch intervals and pitch directions compared to a tone pair. This facilitatory effect of the sequence context was not observed with Monica. The fact that Monica detects rising pitch changes somewhat better than falling pitch changes across contexts also argues against a working memory problem. There is no reason to expect working memory to be tuned to specific pitch directions. In contrast, this peculiar and systematic pitch defect points to the presence of an anomalous pitch perceptual system.

We construe that this striking pitch-processing deficit may well account for the music-specific disorder observed in Monica. In effect, Monica is unable to perceive pitch variations that lie at a semitone distance, which corresponds to 1/12 of an octave and is the smallest pitch interval that is used in Western scales. Normal discrimination abilities in the large majority of adult and infant subjects are more fine grained, on the order of 1/4 tone, in both ascending and descending directions (Olsho et al., 1982). In contrast, for Monica, most musical pitch variations lie below threshold.

Is Speech Intonation Spared?

Since Monica suffers from a pitch discrimination deficit that is remarkably severe, she might suffer from an impairment in the discrimination of speech intonation patterns on the basis of pitch cues alone. To assess this possibility, we exploited once again experimental tests that have been previously used with brain-damaged amusic patients (Patel et al., 1998). These tests are constructed by computer editing two basic sets of sentences that only differ from each other 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 rise is on the order of 3–11 semitones for questions, while the pitch fall is of 2–3 semitones for statements. In a second set of sentences, the pitch difference (of a magnitude of eight semitones) affects an internal word of the sentence to mark emphatic stress, like in "SING now please!" and "Sing NOW please!" (capitals indicate the stressed word).

The two sets of sentences were presented in isolation. Subjects judged whether the sentence indicates a statement or a question for the first set of sentences differing by their final pitch change; or they indicated which word bore the stress for the second set of sentences. These two tasks were relatively easy to complete for both Monica and controls who obtain 100% and 98% correct, respectively, in the final pitch change condition, and 77% and 87% (range: 67–100), respectively, in the internal pitch change condition. Thus, Monica's disorder in monitoring pitch variations does not appear to encompass speech intonation.

Discussion

From the data presented, we conclude that congenital amusia, or tone-deafness, is not a myth (Kazez, 1985),

but a genuine and specific learning disability for music. Affected individuals, who are otherwise unimpaired, have extreme difficulties appreciating, perceiving, and memorizing music. The systematic evaluation of Monica, who reported to be severely handicapped in the musical domain despite her efforts to learn it, largely confirms the presence of an underdeveloped system for processing music. Her musical impairment cannot be explained by hearing loss since she has normal audiometry and is proficient in the recognition of other auditory nonmusical material, such as voices, spoken lyrics, and common environmental sounds. Her musical disorder cannot be explained by a lack of exposure since she had music lessons during childhood and was raised in a family in which a few siblings are musically normal. Finally, the musical deficit cannot be ascribed to some general cognitive slowing, since she scores normally on cognitive tests and has reached a high level of education. The musical disorder appears as an isolated deficit in an otherwise fully normal cognitive and affective system.

Monica's learning disability may arise from a basic problem in pitch discrimination. For Monica, music must sound monotonous since she cannot detect pitch variations that are smaller than two semitones. Thus, she cannot perceive the pitch differences, the tone and semitone, that are the building blocks of the musical scales of most cultures. This pitch defect is construed as the origin of congenital amusia.

Reducing congenital amusia to a single deficient mechanism in pitch perception requires elaboration. First, ascribing musical impairments to a low-level pitch discrimination problem is not trivial. Acquired amusia resulting from brain lesions usually arises from failures specific to higher-level aspects of music processing, by degrading tonal knowledge for example, and is not necessarily associated with a pitch discrimination defect (e.g., Peretz, 1993). Conversely, brain lesions that disturb pitch discrimination do not necessarily affect music processing to the same degree as seen in Monica (Tramo et al., 1990). For instance, excision of cortical tissue within the right Heschl's gyrus produces an elevation in the threshold for discriminating pitch direction (Johnsrude et al., 2000) on the order of two semitones, which is better than Monica's performance. Such lesions do cause perceptual and memory disturbances for melodies, but patients with such lesions cannot be called amusic since they can discriminate above chance (Zatorre, 1985) and can also learn new melodies (Samson and Zatorre, 1992), albeit not as well as controls.

Second, the fine-grained pitch discrimination disorder was not the only impaired musical ability. Monica was also impaired in discriminating melodies by their rhythm. Her difficulty with temporal patterns seems to hold even when all pitch variations are removed, although the nature and extent of this impairment could not be documented. There are several possible explanations for the presence of this myriad of musical deficits. The explanation that we presently favor is that the ensemble of musical deficits are cascade effects of a faulty pitch-processing system. In effect, in our ongoing study of other congenital amusic participants, we have observed that they systematically fail on tests that probe their ability to process musical pitch variations, whereas most of them are able to process musical temporal variations as normal listeners. Thus, the presence of a pitch defect appears diagnostic of the existence of a music-learning disability, whereas the association of a rhythmic problem appears optional. Hence, we construe that a faulty perception of pitch might bring the development of the entire musical system to a halt. In this perspective, fine-grained pitch perception might be an essential component around which the musical system develops in a normal brain.

Further investigation of multiple single cases such as this one, and the joint study of musical pitch and time structure in infants, will be necessary to test the above account. In turn, this research endeavor should provide serious indications as to which processing component is crucial for normal musical development and in what way music might be special. More generally, knowledge of every aspect of this music-specific disorder should enrich current views of other forms of learning disabilities. For instance, one may consider congenital amusia as a mirror image of some developmental disorders of language, whereby pitch would be to music what time is to speech.

Experimental Procedures

Battery of Musical Tests

The musical battery uses a pool of 30 *novel* two-phrase monodies that obey the rules of the Western tonal system. Half were written in a duple meter, half in a triple meter. The two-phrase sequences were used in the metric subtest. For all the other subtests, only the second phrase of each sequence served as a stimulus. These second phrases were 4 bars long, lasted about 4 s, and contained from 8 to 19 tones (mean = 10.7). The stimuli were generated on a computer controlling a Yamaha TX-81Z synthesizer. The chosen tempo was fixed at 120 crochets per minute, and the voice was the approximation of a piano sound. The analog output was recorded on a digital DAT SONY recorder, which was also used to play melodies to the subjects.

In the *contour and interval subtests*, two types of manipulation were applied to the second phrase of 15 original sequences. These manipulations consisted of creating a contour-violated alternate melody by modifying the pitch direction of one tone and an interval-violated alternate melody by modifying the pitch of the same critical tone to the same extent in terms of semitone distance but in keeping with the original contour. The changed pitch remained in the key of the melody. Its serial position varied across melodies; half fell in the beginning of the melody and half fell in the end, while avoiding the first and last tone positions. Average pitch interval changes were 4.3 and 4.2 semitones apart from the original pitch in the contour-violated and interval-violated condition, respectively. The minimal pitch change was set to three semitones, and the maximal pitch change was set to seven semitones. The changes generally fell into the frequency range of the melody. Two sets, each consisting of 2 practice trials and 30 experimental trials, were constructed with these melodies. Each trial consisted of a warning signal and a target melody followed, after a 2-s silent interval, by a comparison melody. Duration of the intertrial interval was 5 s long. Each set consisted of 15 pairs that were made of identical melodies and 15 trials of different melodies, presented in a random order.

As mentioned previously, in the *metric test*, the 30 entire two-phrase sequences served as stimuli. Each sequence was recorded in a random order with an intertrial interval of 5 s. These experimental trials were preceded by four practice trials. For the *rhythmic test*, the stimuli were derived from the second phrase of the 30 sequences used in the metric task, in order to correspond to the target melodies used in the contour and interval subtests. One manipulation was applied to these isolated phrases to create different comparison patterns. These temporal alternates involved a temporal grouping

change by changing the durational values of two adjacent tones. This local change was such that the size of each temporal group defined by temporal proximity was changed, while keeping the meter and the total number of sounds identical. The serial positions of these changes varied across patterns. Thus, the only cue available for discrimination was the temporal grouping of the tones (i.e., the rhythm). A set of 2 practice and 30 experimental trials was constructed with these temporal patterns in the same way as the contour and interval conditions.

For the contour and interval subtests and the rhythmic test, subjects were required to perform a "same-different" classification task. They had to judge, in each trial, whether the target sequence and the comparison sequence were the same or not. Prior to each condition, two practice trials were presented. For the metric task, subjects were informed that they would be hearing waltzes and marches, which they had to discriminate along that dimension. Subjects were encouraged to tap along with what they perceived to be the underlying beat of each sequence. Feedback on the response was only provided in the practice trials.

Psychophysical Tests of Pitch Discrimination

A standard, psychophysical forced-choice procedure was used in successive blocks of 40 trials each, starting with the largest pitch distance. In each block, half the stimuli contained no change and half contained a rising or falling pitch change; trials were presented in a random order. Monica was first tested with a five-tone sequence in which the fourth tone could vary in pitch. We started with the largest changes of 11 semitones and with piano tones of different durations. The following blocks used smaller pitch distances and pure tones of 700-ms duration only since these tones were reported to be intelligible by Monica. In a subsequent session, Monica was presented with the same pure 700-ms tones, but within a pair of stimuli such that the second tone was either the same or different than the first. Monica was invited to press a "yes" key when she detected a change and to press a "no" key when she detected no change. Feedback was provided on the screen of the computer, and the task was self paced.

All stimuli were generated digitally using the MITSYN signal-processing software at a sampling rate of 44.1 kHz. A sequence consisted of five successive tones, and a pair consisted of two successive tones. The duration of each tone was 700 ms, unless noted otherwise, with the sound envelope of a piano tone. The frequency spectrum of each tone either corresponded to the one of a piano tone or to a single frequency with a similar amplitude counter (referred to here as "pure tones"). All target tones corresponded to C5 (with a fundamental frequency of 524 Hz). In half the stimuli, either the fourth tone of the sequence or the second tone of the pair could take a different pitch value. These different pitch values correspond to B5 (988 Hz) or C4 sharp (277 Hz) for the 11-semitone distance, to E5 (660 Hz) or A4 flat (416 Hz), D5 sharp (623 Hz) or A4 (440 Hz), D5 (588 Hz) or B4 flat (466 Hz), and C5 sharp (555 Hz) or B4 (494 Hz) for the four-, three-, two-, and one-semitone distance, respectively. The stimuli were presented bilaterally through Sennheiser HD450 headphones, in a quiet room, at an intensity level of 70 dB SPL-A, via the Media Control Functions program.

Pitch Variation Discrimination in Speech

A total of 24 sentence pairs naturally spoken by a native female French speaker were recorded. Each pair represented two lexically identical versions of a sentence but differed in intonation. A set of 12 pairs constituted statement-question pairs (e.g., "Il parle Français." versus "Il parle Français?"). The other set of 12 pairs constituted focus-shift pairs, involving a shift in the word that bore the focus of the sentence (e.g., "Allez DEVANT la banque, j'ai dit." versus "Allez devant la BANQUE, j'ai dit."; capitals indicate stress). All pairs were acoustically adjusted by computer editing to equalize patterns of syllable timing and loudness within each pair, yielding natural-sounding sentence pairs in which pitch variations were the only salient cue for intonation interpretation. The computer-editing procedure is described in detail in Patel et al. (1998). The sentences were presented one at a time in a random order. For the final pitch change sentences, the task of the subject was to judge whether the utterance was a question or an assertion. For the internal pitch change sentences, the task of the subject was to indicate which word bore the stress. Each task was performed on 32 trials. No feedback was provided to the subject.

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