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Research report

Congenital amusia in childhood: A case study

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ABSTRACT

Here we describe the first documented case of congenital amusia in childhood. AS is a 10-year-old girl who was referred to us by her choir director for persisting difficulties in singing. We tested her with the child version of the Montreal Battery for the Evaluation of Amusia (MBEA) which confirmed AS's severe problems with melodic and rhythmic discrimination and memory for melodies. The disorder appears to be limited to music since her audiometry as well as her intellectual and language skills are normal. Furthermore, the musical disorder is associated to a severe deficit in detecting small pitch changes. The electrical brain responses point to an anomaly in the early stages of auditory processing, such as reflected by an abnormal mismatch negativity (MMN) response to small pitch changes. In singing, AS makes more pitch than time errors. Thus, despite frequent and regular musical practice, AS's profile is similar to the adult form of congenital amusia.

1. Introduction

Congenital amusia is a disorder preventing afflicted individuals from perceiving music normally. It is currently diagnosed with the help of the Montreal Battery of Evaluation of Amusia (MBEA; Peretz et al., 2003). This battery distinguishes between people with congenital amusia and the general population by assessing the discrimination of melody, rhythm and meter, as well as incidental memory for short musical excerpts. Amusia is defined as a severe impairment in most of these tests, particularly in the discrimination of pitch variations (Peretz, 2008).

Congenital amusia may originate from a deficit in fine-grained pitch perception (Foxton et al., 2004; Hyde and Peretz, 2004; Peretz et al., 2002). For example, amusic adults cannot detect pitch changes smaller than a semitone in oddball tasks, and show no sign of improvement with practice, as opposed to normal adults (Hyde and Peretz, 2004). In contrast, in a similar context, amusics can detect time changes as well as controls (Hyde and Peretz, 2004). The pitch discrimination deficit found in amusia can be tied to abnormal electrical brain responses, as reflected by the absence of a normal P300 (Peretz et al., 2005). Yet, amusics seem to exhibit normal early automatic brain responses, by displaying a normal mismatch negativity (MMN) even for pitch deviations of half a semitone (Moreau et al., 2009; Peretz et al., 2009). These results suggest that amusic adults' auditory cortices have developed normally and that the problem underlying the pitch deficit occurs later

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along the auditory pathway, as the perception of pitch differences reaches awareness.

This pitch deficit may also manifest itself in poor singing (Dalla Bella et al., 2009). Most amusics sing out of tune by making anomalously high number of pitch interval and contour errors. Rhythm seems less impaired, with more than half of the amusics singing in-time.

In sum, in adults, congenital amusia is characterized by impaired pitch discrimination abilities that may compromise music perception and production. Until now, this disorder has been documented in the adult population only. Considering that adult amusics report musical failures as far as they can remember and that a number of key musical skills are acquired before the age of six in normal development (Hannon and Trainor, 2007), one would expect to find evidence of amusia in school-age children. Here we report such a case.

2. Case history

AS is a left-handed 10-year-old girl who was referred to us by her choir director for persisting difficulties in singing. In more than 30 years of career, the choir director has never encountered such an enduring difficulty in helping a child improve her musical skills despite considerable efforts invested by both parties.

AS was evaluated at the end of fourth grade at elementary school. She is an only-child, shy tempered, whose first language is Russian; she learned French upon arrival in Quebec, Canada, at the age of six and a half. She speaks French at school but not at home.

When AS lived in Moscow, she had group music lessons in kindergarten and later, group music lessons in elementary school in Quebec. Her mother, who highly values music, used to play Mozart recordings frequently and in different situations (during meals, at bed time, etc.) as AS was growing up. Ever since they moved to Canada, AS and her mother attended numerous classical music concerts. AS attends approximately 10–12 concerts/year. Above all, AS has been actively involved in a choir since September 2008 (about 20 months prior to testing), which requires her to take part in 2-hour practice sessions twice a week and four concerts a year.

AS was born without complications at the end of a normal pregnancy. Her weight at birth as well as the development of language and motor skills, such as walking, were normal. According to her mother, she has never contracted any serious disease and has never had any brain injury.

At school, AS performs in the normal range when compared to her classmates in English (AS = 90%, group average = 79%), Mathematics (AS = 79%, group average = 79%), French (AS = 75%, group average = 78%), even though it is not her first language, and Arts classes (AS = 80%, group average = 80%). Surprisingly, AS's results in music are also in the average range (AS = 84%, average = 84%).

In the present study, we compared AS with three controls (CP, GBT, MA) matched for gender, age and schooling (see Table 1). Like AS, the matched controls took part in group music lessons at school. In addition, two of them had just started singing in extra-curricular activities (choir rehearsals once a week for CP, and private singing lessons once a week for GBT) 2 months before testing. Thus, AS has more musical experience than her matched controls. The fact that AS is left-handed, while the controls are right-handed, may not account for her lower musical scores, as group studies indicate that left-handers perform better than right-handers in pitch discrimination (Deutsch, 1978; Deutsch, 1980), memory for tonal sequences and timbre recognition (Byrne and Sinclair, 1979).

Table 1 – Characteristics of children and obtained scores for the child version of the MBEA (short version) for the amusic case –AS (in bold) – and her three matched controls (CP, GBT, MA), along with a further group of 20 children for comparison.

	AS	CP	GBT	MA	Comparison group
Gender	**F**	F	F	F	14 F; 6M
Age	**10**	10	10	10	10
Education (grade)	**4**	4	4	4	4
Handedness	**L**	R	R	R	18R; 2L
Melody, total (/20)	**7**	19	17	18	17.2
Scale (/3)	**0**	3	2	2	
Contour (/4)	**2**	4	4	4	
Intervals (/3)	**1**	2	2	2	
Rhythm (/20)	**10**	20	20	17	19.0
Memory (/20)	**12**	18	18	17	17.4
Composite score (/60)	**29**	57	55	52	53.4

3. General auditory and cognitive abilities

3.1. *Audiometry*

AS's hearing level is normal for her age group with thresholds below or at 20 dB HL (Cunningham and Cox, 2003; see Table 2).

3.2. *Intellectual functions*

AS's intelligence score is average. Her intellectual global score is 102, as measured by the Wechsler Intelligence Scale for Children, fourth edition (WISC-IV; Wechsler, 2003), which corresponds to the 55th percentile of her age group. Her profile is homogeneous, with average perceptual (39th percentile), verbal (36th percentile), working memory (58th percentile) and processing speed abilities (27th percentile). These results might be slightly under-estimated because AS was evaluated in her second language (French). Nevertheless, the WISC-IV results globally match her school performance.

Table 2 – AS' Thresholds in audiometry for each ear and frequency.

	Left ear (dB HL)	Right ear (dB HL)
.5 KHz	10	10
1 KHz	5	0
2 KHz	0	0
3 KHz	5	0
4 KHz	0	0
6 KHz	20	10
8 KHz	20	5

3.3. Language

AS's language abilities are also normal. Her performance on the French version of the Expressive One-Word Picture Vocabulary Test (EOWPVT; Brownell, 2000), commonly used to evaluate expressive language in children, is average (Standard score: 95, 37th percentile). She also completed the French version of the Peabody Picture Vocabulary Test (PPVT) – Revised (Dunn et al., 1993), which is frequently used to evaluate receptive language abilities. AS's results on this task are in the low average, with a standard score of 86 (16th percentile). Even though lower than the others, this score is not alarming since it is still in the normal range and could be explained by the normal intra-individual variability commonly found in any neuropsychological evaluation. Moreover, it is important to remember that French is a recently acquired language for AS, as she has been living in Quebec, Canada, for only three and a half years, and that she only speaks French in school. Her verbal abilities are therefore considered normal, as estimated by the WISC-IV, her average marks in French at school and her performance on the receptive and expressive language tests, all evaluated in her second language.

3.4. Attention

AS has very good attention capacities, according to her results on the French version of the Test of Every Day Attention for Children (TEA-CH) (Manly et al., 2006). She completed different tasks evaluating visual attention, auditory attention and divided attention (i.e., completing two tasks at the same time). Her results on these tasks were in the average range or above (*Recherche dans le ciel*, visual attention score = 55th percentile; *Coups de fusils,* auditory attention score = 100th percentile; *Faire deux choses à la fois,* divided attention = 35th percentile).

4. Musical tests: results and comments

4.1. Is AS amusic?

In order to assess AS's musical abilities, we used the reduced child version of the MBEA. Like in the adult version of the MBEA, the tasks are quite simple. Children are asked to compare a target melody to a standard melody and to decide if they are the same or different. They also have to complete a yes–no recognition task in the memory test. However, the adult version of the MBEA comprises six tests (180 trials) and takes about 1.5 h to complete for a normal adult. This duration far exceeds the average attention capacity of a child. To make the MBEA more child-friendly, we adjusted it in two steps. In the first step, we reduced both the length of the melodies to be discriminated (mean duration: 3.5 sec; mean number of notes: 7.1) and the number of trials per test (20 trials, 10 different). In a second step, we reduced the five tests (scale, contour, interval, rhythm and memory) to three tests only, assessing the discrimination of melodic changes (scale, contour and intervals) in a single melody test, rhythm discrimination and memory. This reduced version has been assessed and validated in 86 children residing in Montreal (see www.brams.umontreal.ca/plab/publications for both the battery and norms).

AS's musical perception was evaluated with the short version of the MBEA for children, involving the three tests that evaluate melody discrimination, rhythm discrimination and memory, in less than 25 minutes. The melodies have been adapted from the MBEA for adults (see Fig. 1) and are presented with different timbres (Pizzicato violin, Trumpet, Marimba, Vibraphone, Flute, Guitar, Oboe, Harp, Clarinet, and Piano). The melody timbre is kept constant in a given trial. In the melody test, 10 pairs of stimuli are made of identical melodies and 10 pairs are made of slightly different melodies. When different, the second melody of the pair contains either (a) a key change by modifying one note of the melody by another note that is out-of-key in three trials; (b) a contour change by changing the pitch direction of one note while maintaining the key in four trials; and (c) an interval change by changing one note while maintaining the same contour and key in three trials. The rhythm discrimination task also consists of 20 pairs of melodies, in which half of the trials contain a rhythmic modification in changing the duration values of two adjacent tones while keeping meter and number of notes unchanged. An example of stimuli is presented in Fig. 1. For the two tests of melody and rhythm discrimination, the participant has to decide if the two melodies are the same or different. The third test evaluates incidental memory for music. Among the 20 stimuli of the task, 10 are novel melodies, and 10 are melodies that were presented in the first two tests. For each item of this memory task, the participant has to decide if the melody was presented before or not. The participants had to write their answers on a sheet with symbols as shown in Fig. 1 (as is the case in the adult version of the MBEA). The stimuli were presented through speakers on a personal computer with the software Windows Media Player.

AS's performance on the MBEA is at chance (see Table 1). Her global score averaged across the three tests is 48% of correct answers (Hits, indicating the percentage of "different" responses to a different melody": 46.7%, False alarms, reflecting "different" responses to identical trials: 50%), which is clearly impaired as compared to the three control subjects (CP, GBT, and MA; Table 1). The control subjects' performance (mean: 91.3%; range: 86.7–95) is similar to that of a group of 20 children with no established musical deficiencies (fourteen 10-year-old girls and six 10-year-old boys) with a global score of 89.3% [standard deviation (SD) = 3.9]. On the melody

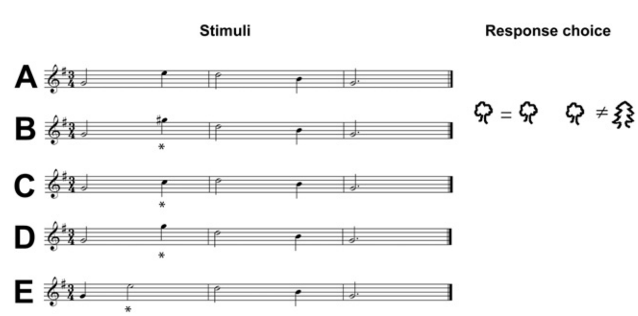

Fig. 1 – Example of a stimulus used in the child version of the MBEA. The original stimulus is presented in (A) its scale alternate in (B) its contour alternate in (C), its interval alternate in (D) and its rhythm alternate in (E). Asterisks indicate the changed note. The symbols provided on the response sheet are presented on the right.

discrimination test, AS's score was below those of controls (90%, range: 85–95, and 86.1%, SD = 9.5 for the three matched controls and the other 20 normal children, respectively) with 35% of correct answers (Hits: 30%, False Alarms: 60%). On the rhythm discrimination test, AS also performed at chance level, with 50% of correct answers (Hits: 50%, False-Alarms: 50%), while the controls obtained scores of 95.0% (range: 85–100), and 95.3% (SD = 4.9). On the memory test, AS's score was 60% correct (Hits: 60%, False Alarms: 40%) while the matched controls and normal children obtained scores of 88.3% (range: 85–90) and 86.7% (SD = 7.5) respectively. Thus, compared to children of her age, AS shows a severe impairment in music discrimination and recognition.

4.2. *Is the musical impairment due to poor pitch discrimination?*

AS's pitch discrimination was evaluated through an oddball task similar to the one previously used with adult amusics (Hyde and Peretz, 2004). She was presented with five tone sequences and was requested to evaluate if the fourth tone differed in pitch when compared to the other four tones. She had to press the "change" key if she detected a pitch difference and the "no change" key if she did not. Two-hundred and forty sequences were presented. Half the sequences comprised five identical repetitive tones (i.e., the standard) and half included a fourth pitch-displaced tone (by 25 or 200 cents up or down, 100 cents corresponding to one semitone). The tones were 100 msec long synthesized piano notes and the standard tone corresponded to C6 (1047 Hz). Within a sequence, the inter-stimulus interval was 400 msec and a 2 sec answer time period was allowed. In order to make sure that the participants understood the task and were comfortable with the material, they were given 10 practice trials (two standards, two 200-cent-up differences, two 200-cent-down differences, two 25-cent-up differences, and two 25-cent-down differences, in this order). If the child did not seem to understand the task, the practice trials could be repeated. Neither AS nor the controls needed more than two series of practice trials.

The results were calculated in terms of hits and false alarms. A hit was scored when the child pressed the "change" button when a change was present (either 25 or 200 cents) in the five-tone sequence. A false alarm was scored when the child pressed the "change" button when no change was present. The results indicate that AS's fine-grained pitch discrimination is impaired (see Table 3). For the 25 cent pitch differences, she performed at chance level. In contrast, for the 200 cent pitch differences, her performance was normal. Thus, AS's musical difficulties seem to be associated with a deficit in fine-grained pitch discrimination, as observed in the amusic adult population (Hyde and Peretz, 2004; Peretz et al., 2005).

4.3. *AS's brain responses*

AS's electrical brain responses to sounds (presented binaurally in headphones; 70 dB SPL) were recorded while she was instructed to ignore them and to watch a self-selected silent subtitled movie. Her brain responses were compared to those of five controls, one boy and four girls, of approximately the same age (mean = 11.3 years). The stimuli were the same

Table 3 – Percentages of hits and false alarms (F.A.) in the detection of 25 and 200 cent changes in the amusic case – AS (in bold) and her three matched controls (CP, GBT, MA).

		AS	CP	GBT	MA
25 cents	Hits	**13.3%**	100%	100.0%	98.4%
	F.A.	**6.7%**	7.5%	1.7%	2.5%
200 cents	Hits	**96.7%**	98.33%	100.0%	98,3%
	F.A.	**6.7%**	7.5%	1.7%	2.5%

tones as used in the pitch discrimination task but, instead of being organized in five tones sequences, they were presented in a continuous sequence with a tone onset to onset interval of 500 msec, for a total of 3040 tones, with 90% of the tones being standard and 10% being pitch deviant tones (with 152 deviations of 25 cents and 152 deviations of 200 cents).

AS's electroencephalography (EEG) responses were recorded using a 66 channel cap with a Neuroscan system (Compumedics Neuroscan, El Paso, Tx) at a recording frequency of 250 Hz. After the recording session, the data were corrected for eye movement and epoched in 600 msec windows. A baseline correction with a time window of 100 msec prior to stimulus onset and an artifact rejection (−100 to 100 μV) were conducted. As a result of the artifact rejection, for AS, 11% of the 25 cent deviant tones and 13% of the 200 cent deviant tones were rejected. In controls, 21% of the 25 cent and 22% of the 200 cent deviant tones were rejected, on average.

Here, we focus on the MMN which is an event-related potential (ERP) component that usually peaks between 100 msec and 250 msec and that is known to reflect pre-attentive change detection (Näätänen, 1992). As can be seen in Fig. 2, a MMN is observed in AS for the 200 cent deviations but not for the 25 cent deviations, whereas a MMN can be observed for both pitch deviations in the control group. AS's MMN amplitude was −4.1 μV with a latency of 224 msec for the 200 cent deviations, which is comparable to the MMN of the controls (with an average amplitude of −3.9 μV; range: −2.99 μV to −4.95 μV, and a mean latency of 197 msec; range: 160–200 msec). For the 25 cent deviations, there was no measurable ERP in AS, whereas in the controls, the MMN amplitude was −2.85 μV (range: −.40 μV to −6.033 μV) with a latency of 285 msec (range: 228–340 msec). The smaller amplitude and delayed peak of the normal MMN for 25 cents changes relative to the 200 cents changes reflect a difference in discriminability between the deviants (e.g., Näätänen and Picton, 1987).

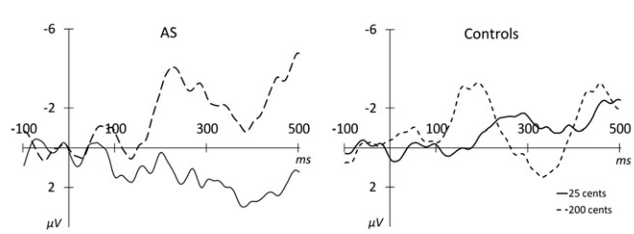

Fig. 2 – MMN subtracted waveforms (deviant minus standard) recorded at Fz for the amusic case – AS-and five matched controls.

4.4. *Is singing impaired?*

AS sang two songs from memory. She chose “Frère Jacques” (the French version of “Brother John”) and “Bonne Fête” (the French version of “Happy Birthday”). “Frère Jacques” is composed of 32 notes (31 intervals) and has a range of 14 semitones. “Bonne Fête” contains 26 notes (25 intervals) and its range is of one octave (12 semitones). We asked the three previously presented matched controls (CP, GBT, and MA) to sing the same two songs.

AS’s and controls’ productions were recorded in a quiet environment in the presence of the experimenter only. A personal computer, a Sure microphone, and the Adobe Audition 1.0 software were used to record the productions. The software Praat as well as scripts developed for another study (Hébert et al., 2008) were used for the acoustical analysis of the songs. The pitch tracking function of the software Praat was used to calculate the mean frequency of sung tones. Pitch interval and contour errors were then compiled. For the rhythm dimension, the program calculated inter-onset intervals (IOIs) which allowed the identification of rhythm errors.

The self-chosen tempi were 171 beats/min (bpm) for AS, 129 for CP, 125 for GBT and 171 for MA for the song “Frère Jacques”. AS sang “Bonne Fête” at a tempo of 111 bpm, while CP, GBT, and MA sang it at tempi of 129, 138, 108 bpm, respectively. Since AS omitted to sing the first six notes of “Bonne Fête”, these were excluded from the data analyses in all four participants. Results are averaged across the two songs.

4.4.1. *Pitch*

As no starting pitch was imposed to the participants, their singing was analyzed in a relative way by measuring intervals instead of absolute pitch heights. Sung intervals were compared to target intervals as prescribed by the musical notation of the songs. An interval was considered as erroneous when its size was one semitone larger or smaller than the target interval (as in Dalla Bella et al., 2007). The number of interval errors was compiled. In addition, in order to examine if the interval errors were in- or out-of-key, the mean frequencies of the produced tones calculated via the software Praat (as in Hébert et al., 2008) were approximated to the closest notes of the chromatic scale. The sung melody’s key was then inferred from the starting pitch and each erroneous note (i.e., presenting an interval error) was evaluated as being part of the corresponding key or not (as in Dalla Bella et al., 2009). Finally, the average size of the interval deviations (i.e., the average distance between the sung and the target interval in semitones) was also calculated.

Out of the 50 pitch intervals sung, AS made 20 interval errors whereas CP, GBT, and MA made respectively 14, 1, and 11 errors. The number of out-of-key interval errors made by AS is comparable to the controls’ (Table 4). AS also produced larger pitch deviations than the controls, with an average of 1.5 semitones as compared to .6 for the controls (d of Cohen = .86, Effect size = .4). When examining deviations as a function of interval size (intervals of 0, 2, 3, 4, 5, 7, 8 and 12 semitones), we noted that AS is more impaired on large than on small pitch intervals. There was a significant correlation between the size of the target interval and the size of the pitch deviation for AS ($r = .90$; $p < .01$), but not for the controls ($r = .32$, *n.s.*). Thus, AS seems more affected than controls by the size of the target interval.

Table 4 – Acoustical analysis of singing in the amusic case –AS (in bold) – and her three matched controls (CP, GBT, MA).

	AS	CP	GBT	MA
Number of interval errors	**20**	14	1	11
Number of out-of-key errors	**9**	3	1	8
Interval deviation (semitones)	**1.5**	.6	.3	.8
Number of contour errors	**3**	1	0	3
Number of time errors	**6**	0	3	3
Time deviations (%)	**14.2%**	9.6%	12.3%	9.5%
Temporal variability index	**.19**	.15	.17	.15

4.4.2. *Contour*

Pitch direction was considered as ascending or descending when the interval was larger than one semitone. A contour error was counted when the produced and the target interval direction differed. AS did not make more contour errors than the controls. She produced 3 contour errors whereas CP, GBT and MA made 1, 0 and 3 errors, respectively. Thus, AS’s contour production is normal.

4.4.3. *Rhythm*

A time error was computed when the sung note’s duration was 25% longer or shorter than its predicted value as prescribed by musical notation. The number of time errors was calculated. Furthermore, temporal variability (coefficient of variation of the quarter-note IOI) was estimated by calculating the quarter notes IOIs and dividing the SD by the mean IOI for each song, thus eliminating the confounding effect of tempo variability among the participants and songs (as in Dalla Bella et al., 2009).

Overall, AS made 6 time errors, whereas CP, GBT and MA made 0, 3 and 3 errors, respectively. AS’s performance on “Bonne Fête” was as good as that of the controls, with only 1 time error (CP = 0; GBT = 1; MA = 1 time errors). Nevertheless, for “Frère Jacques”, AS performed more poorly than the controls, with 5 time errors (CP = 0; GBT = 2; MA = 2 time errors). Furthermore, AS’s production of “Frère Jacques” was slightly more variable than the controls’, with an index of .23 versus .16, .15 and .16, for CP, GBT and MA respectively. For “Bonne Fête”, AS’s temporal variability was similar to that of the controls with an index of .15 (.14, .18 and .14 for CP, GBT and MA respectively). Thus, AS’s rhythm production seems to be slightly affected as compared to the controls, but not nearly as much as pitch production.

5. General discussion

AS’s case study suggests that congenital amusia can be observed in childhood and presents itself in a very similar manner as in amusic adults. AS has severe problems in perceiving, memorizing, and producing music. Since the child has normal audiometric results and does not have any cognitive or language impairment, the deficit seems to be isolated and specific to music. In addition, her amusic profile cannot be explained by lack of exposure or motivation since she was raised in a musically enriched environment and had regular extra-curricular choir practice sessions.

Like in the adult form of amusia, AS exhibits a deficit in fine-grained pitch perception. This deficit may account for the abnormal development of her musical competences considering her age (Peretz, 2008). AS's electrical brain responses also point to an anomaly in detecting small pitch deviations. AS did not display a normal MMN for the 25 cent pitch deviations but did so for the 200 cent deviations. These results suggest that, unlike adults, AS's brain fails to respond to the fine-grained pitch variations which may reflect a delay in maturation of the auditory cortex. Thus, the present ERP results suggest a possible difference between the developing amusic profile and the stable phenotype of amusic adults.

When singing, AS is able to maintain the general contour of the song, without being able to sing its precise intervals. This preserved ability to sing pitch directions associated to impaired discrimination of pitch contour, is consistent with recent studies conducted on amusic adults who were able to reproduce pitch intervals in the correct direction (but not with the correct interval size) while being unable to report whether the interval was going "up" or "down" (Loui et al., 2008; Dalla Bella et al., 2009). This dissociation suggests that some aspects of singing may be spared despite impaired perception.

In sum, AS's congenital amusia is fully expressed and is similar to its adult form despite extensive musical training. In order to address the putative contribution of musical lessons on amusia manifestation, one needs to train a cohort of children at risk of developing congenital amusia. The problem with the recruitment of potentially amusic children is that these cases go unnoticed or are extremely rare. It is worth emphasizing here that AS was diagnosed by a music teacher and later confirmed by objective testing. As it is unlikely that the educational system will identify further amusic children in sufficient numbers, we will need to diagnose them on the basis of their scores on the MBEA (Peretz et al., 2003) or on its online version (Peretz et al., 2008). It remains to be determined if further cases of amusic children can be identified on the basis of the child version of the MBEA and if these poor-performing children will exhibit a similar profile as AS.

To conclude, the current single case study shows that congenital amusia can be uncovered in childhood. To date, AS is the only childhood case of congenital amusia, while there is a rapidly increasing number of amusic adult studies in the literature. Thus, we have as yet little understanding of the developmental trajectory of this apparently common developmental disorder. We hope that our study encourages further research aimed at delineating the profile of the childhood form of congenital amusia and at developing interventions that may improve these children's musical skills.

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REFERENCES

Brownell R. *Expressive One-word Picture Vocabulary Test (EOWVPT)*. *Novato*. California: Academic Therapy Publication, 2000.

Byrne B and Sinclair J. Memory for tonal sequence and timbre: A correlation with familial handedness. *Neuropsychologia*, 17(5): 539–542, 1979.

Cunningham M and Cox EO. Hearing assessment in infants and children: Recommendations beyond neonatal screening. *AAP News/American Academy of Pediatrics*, 111(2): 436–440, 2003.

Dalla Bella S, Giguère J-F, and Peretz I. Singing proficiency in the general population. *The Journal of the Acoustical Society of America*, 121(2): 1182–1189, 2007.

Dalla Bella S, Giguère J-F, and Peretz I. Singing in congenital amusia. *The Journal of the Acoustical Society of America*, 126(1): 414–424, 2009.

Deutsch D. Handedness and memory for tonal pitch. In Herron J (Ed), *Neuropsychology of Lefthandedness*. Academic Press, 1980: 263–272.

Deutsch D. Pitch memory: An advantage for the lefthanded. *Science*, 199: 559–560, 1978.

Dunn LM, Thériault-Whalen CM, and Dunn LM. *Échelle de vocabulaire en images Peabody: Adaptation française du Peabody Picture Vocabulary Test-Revised*. Toronto: PSYCAN, 1993.

Foxton JM, Desn JL, Gee R, Peretz I, and Griffits TD. Characterization of deficits in pitch perception underlying 'tone deafness'. *Brain: A Journal of Neurology*, 127(4): 801–810, 2004.

Hannon EE and Trainor LJ. Music acquisition: Effects of enculturation and formal training on development. *Trends in Cognitive Sciences*, 31(11): 466–472, 2007.

Hébert S, Béland R, Beckett C, Cuddy LL, Peretz I, and Wolforth J. A case study of music and text dyslexia. *Music Perception*, 25(4): 369–381, 2008.

Hyde KL and Peretz I. Brains that are out of tune but in time. *Psychological Science*, 15(5): 56–360, 2004.

Loui P, Guenther F, Mathys C, and Schlaug G. Action perception mismatch in tone-deafness. *Current Biology*, 18(8): R331–R332, 2008.

Manly T, Robertson IH, Anderson V, and Mimmo-Smith I. *Test d'Évaluation de l'Attention Chez l'Enfant*. Paris: ECPA – Editions du Centre de Psychologie Appliquée, 2006.

Moreau P, Jolicoeur P, and Peretz I. Automatic brain responses to pitch changes in congenital amusia. *Annals of the New York Academy of Sciences*, 1169: 191–194, 2009.

Näätänen R. *Attention and Brain Function*. Hillsdale, NJ: Erlbaum, 1992.

Näätänen R and Picton T. The N1 wave of the human electric and magnetic response to sound: A review and analysis of the component structure. *Psychophysiology*, 24(4): 375–425, 1987.

Peretz I. Musical disorders: From behavior to genes. *Current Directions in Psychological Science*, 17(5): 329–333, 2008.

Peretz I, Ayotte J, Zatorre R, Mehler J, Ahad P, Penhune V, and Jutras B. Congenital amusia: A disorder of fine-grained pitch discrimination. *Neuron*, 33: 185–191, 2002.

Peretz I, Brattico E, Järvenpää M, and Tervaniemi M. The amusic brain: In tune, out of key, and unaware. *Brain: A Journal of Neurology*, 132(5): 1277–1286, 2009.

Peretz I, Brattico E, and Tervaniemi M. Abnormal electrical brain responses to pitch in congenital amusia. *Annals of Neurology*, 58(3): 478–482, 2005.

Peretz I, Champod AS, and Hyde K. Varieties of musical disorders: The Montreal battery of evaluation of amusia. *Annals of the New York Academy of Sciences*, 999: 58–75, 2003.

Peretz I, Gosselin N, Tillmann B, Cuddy L, Gagnon B, Trimmer C, Paquette S, and Bouchard B. On-line identification of congenital amusia. *Music Perception*, 25(4): 331–343, 2008.

Wechsler D. *Wechsler Intelligence Scale for Children – Fourth Edition (WISC-IV)*. San Antonio, Texas: The Psychological Corporation, 2003.