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Comorbidity and cognitive overlap between developmental dyslexia and congenital amusia

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ABSTRACT

This study investigated whether there is a co-occurrence between developmental dyslexia and congenital amusia in adults. First, a database of online musical tests on 18,000 participants was analysed. Self-reported dyslexic participants performed significantly lower on melodic skills than matched controls, suggesting a possible link between reading and musical disorders. In order to test this relationship more directly, we evaluated 20 participants diagnosed with dyslexia, 16 participants diagnosed with amusia, and their matched controls, with a whole battery of literacy (reading, fluency, spelling), phonological (verbal working memory, phonological awareness) and musical tests (melody, rhythm and metre perception, incidental memory). Amusia was diagnosed in six (30%) dyslexic participants and reading difficulties were found in four (25%) amusic participants. Thus, the results point to a moderate comorbidity between amusia and dyslexia. Further research will be needed to determine what factors at the neural and/or cognitive levels are responsible for this co-occurrence.

Introduction

Developmental dyslexia and congenital amusia are two neurodevelopmental disorders that affect a particular cognitive domain, namely reading and music perception, despite normal sensory and intellectual functioning and despite opportunities for acquiring the relevant skill. Likewise, these disorders have a genetic component and are associated with reduced fronto-temporal connectivity (Boets et al., 2013; Peretz, 2016). To date, there is no evidence of co-occurrence between dyslexia and amusia. The goal of the present study was to assess the relationship between the two disorders.

Developmental dyslexia is a specific and significant impairment in the development of reading skills that is not solely accounted for by mental age, visual acuity problems, or inadequate schooling (World Health Organization, 2011). About 3%–7% of schooled children are affected (Lindgren, Renzi, & Richman, 1985) and difficulties persist into adulthood. The predominant etiological view of dyslexia is the phonological deficit theory, which postulates that reading difficulties

originate from a cognitive deficit that is specific to the representation and processing of speech sounds (Ramus, 2003; Vellutino, Fletcher, Snowling, & Scanlon, 2004). The phonological deficit is manifested in three main areas: phonological awareness (explicitly attending to, judging, and manipulating speech sounds), verbal short-term and working memory (short-term storage, manipulation, and repetition of words or pseudowords) and rapid lexical retrieval (tested by Rapid Automatized Naming: speeded naming of lists of digits, colours, or objects) (Snowling, 2000; Wagner & Torgesen, 1987). Such a phonological deficit seems to play a causal role in the development of poor reading skills (Lyytinen et al., 2004; Puolakanaho et al., 2007). Visual or visual-attentional deficits have been proposed as an alternative proximal cause of dyslexia (Stein & Walsh, 1997; Valdois, Bosse, & Tainturier, 2004; Vidyasagar & Pammer, 2010), but may affect only a subset of individuals with dyslexia (Ramus et al., 2003; Saksida et al., 2016).

Similarly, congenital amusia refers to lifelong deficits that appear specific to music processing and cannot be

attributed to hearing loss, intellectual deficiencies, or lack of music exposure (Ayotte, Peretz, & Hyde, 2002). The most common form of congenital amusia, which will be the focus here, concerns the conscious processing of the pitch structure of music. Amusic individuals have a normal understanding of speech and prosody in everyday life. They can recognize speakers by their voices and can identify all sorts of familiar environmental sounds such as animal cries. What characterizes them behaviourally is their difficulty with detecting out-of-tune singing, including their own, with recognizing a familiar tune without the aid of the lyrics, and with maintaining short tunes in memory (e.g., Tillmann, Lévêque, Fornoni, Albouy, & Caclin, 2016). Affected individuals represent 1.5%–4% of the general population (Peretz & Vuvan, 2017).

Despite the apparent specificity of the cognitive deficits characterizing each of these two disorders, some similarities between amusia and dyslexia can be highlighted. At a perceptual level, there is a large literature on auditory deficits in dyslexia (Farmer & Klein, 1995), postulating that early disruption of auditory processing may be the underlying cause of the widely observed phonological deficit. A similar kind of rapid auditory information processing deficit has recently been postulated for amusia (Albouy, Cousineau, Caclin, Tillmann, & Peretz, 2016). Yet, only a subset of dyslexic individuals seem to be affected, and the claim that auditory deficits actually explain the phonological deficit remains controversial to this day (Bishop, 2006; Goswami, 2006, 2014; Ramus, White, & Frith, 2006; Rosen, 2003; Tallal, 2006; White et al., 2006). At any rate, proponents of auditory theories have not claimed or tested a lack of musical skills in dyslexia, but some have proposed to use musical training to remediate auditory, language and reading abilities (Kraus & Chandrasekaran, 2010; Schön & Tillmann, 2015; Tallal & Gaab, 2006). Still, a handful of studies have reported abnormal musical abilities in dyslexia: namely, abnormal musical rhythm perception and production (Huss, Verney, Fosker, Mead, & Goswami, 2011; Overy, Nicolson, Fawcett, & Clarke, 2003; Thomson, Fryer, Maltby, & Goswami, 2006), poor pitch discrimination (Santos, Joly-Pottuz, Moreno, Habib, & Besson, 2007; Ziegler, Pech-Georgel, George, & Foxton, 2012) and reduced working memory for pitch (Weiss, Granot, & Ahissar, 2014).

Although there is no reported evidence of reading impairments in congenital amusia, subtle deficits in processing speech intonation (Hutchins, Gosselin, & Peretz, 2010; Hutchins & Peretz, 2012; Liu, Patel, Fourcin, & Stewart, 2010; Patel, Wong, Foxton, Lochy, & Peretz, 2008) and in processing pitch contrasts in tone language words (e.g., for native Mandarin speakers: Nan, Sun, & Peretz, 2010; Tang et al., 2018; Zhang, Peng, Shao, & Wang, 2017; for native French speakers: Tillmann et al., 2011) have been observed. Furthermore, a few studies have reported phonological impairments (Jones, Lucker, Zalewski, Brewer, & Drayna, 2009; Loui, Kroog, Zuk, Winner, & Schlaug, 2011) in amusia. Sun and collaborators (Sun, Lu, Ho, & Thompson, 2017) reported that some amusic individuals with poor pitch discrimination abilities exhibited impairments in phonological awareness, yet were normal in phonological short-term memory or rapid naming. The authors ascribe the pitch and phonological problems to a common origin, namely a defective early acoustic stage of processing. However, it is unclear how an early acoustic defect could impact phonological awareness selectively and why the majority (75%) of amusic cases have no phonological difficulties. Hence, this association between amusia and phonological awareness seems weak and may reflect the contribution of other factors than phonology, such as poor auditory awareness in general. Indeed, both dyslexia and amusia have also been characterized as awareness disorders (e.g., Peretz, Brattico, Järvenpää, & Tervaniemi, 2009; Ramus & Szenkovits, 2008).

In dyslexia, it has been suggested that the phonological deficit might have more to do with perceptual awareness, attention, working memory and task difficulty factors other than the nature of phonological representations per se (Ramus & Szenkovits, 2008). This view is now being supported by a number of studies (Boets et al., 2013; Ramus, 2014; Ramus & Ahissar, 2012). Similarly, the core deficit in congenital amusia has been proposed to reside in a lack of conscious access to processed pitch deviances. The amusic brain can track and record subtle pitch variations as normal individuals do, but the outcome of these computations does not give rise to any conscious report (e.g., Zendel, Lagrois, Robitaille, & Peretz, 2015). This dissociation, or disconnection, can be observed at all levels of processing in amusics, from acoustical analysis to memory representation in both perception and production of music (Peretz, 2016). A related proposal is that the deficit would lie

in pitch short-term memory rather than in pitch representation per se (Tillmann et al., 2016).

Overall, dyslexia and amusia may arise from similar causal chains between genetic variations, perturbations of brain development and the appearance of a deficit at a cognitive level (Galaburda, LoTurco, Ramus, Fitch, & Rosen, 2006; Peretz, 2008; Vellutino et al., 2004). According to a previous aetiological model of developmental disorders (Ramus, 2004), they could be caused by similar neurodevelopmental mechanisms, which would increase their comorbidity. A common primary cause could be the formation of cortical anomalies in specific areas (left perisylvian cortex for dyslexia; right inferior frontal gyrus and auditory cortex for amusia) during neuronal migration, as supported by anatomical abnormalities in both dyslexic and amusic individuals (Galaburda, Sherman, Rosen, Aboitiz, & Geschwind, 1985; Hyde et al., 2007).

The similarities between dyslexia and amusia at perceptual, cognitive, and anatomical levels raise the possibility that a common factor is at play in the emergence of these neurodevelopmental disorders, albeit in different domains. Thus, we investigated to what extent dyslexia and amusia can co-occur in the same individual.

We did so in two distinct studies, with different advantages and limitations. Firstly, we took advantage of a large existing database of online tests and questionnaires taken by an unselected population, which allowed us to investigate potential links between self-reported dyslexia and poor musical performance on on-line assessments of melody and rhythm. Secondly, we carried out a new study comparing two groups of participants diagnosed with either dyslexia or amusia, with a whole battery of literacy and musical tests.

Study 1: music performance in a large pool of self-reported dyslexic adults (*n* = 266)

Peretz and Vuvan (2017) presented the first population-based screen for congenital amusia, with a sample of 18,000 participants. On the basis of three objective tests of musical ability and a questionnaire including self-report of other disorders, they found that amusia emerged in relative isolation from other cognitive disorders. The greatest comorbidity was found for spatial orientation disorder (with 15% in amusia compared to 9.1% in controls). Only 7.7% of people with amusia also reported dyslexia, compared to 6.9% in non-amusic controls. Here, we focus instead on individuals reporting dyslexia and analyse their musical performance, compared to matched individuals who did not report having dyslexia. It may be the case that vulnerability to dyslexia leads to a higher risk of developing amusia than vice-versa.

Methods

Participants

Participants volunteered to test their musical abilities via Brams laboratory's website (http://www.brams.umontreal.ca/online-test/) between July 2008 and February 2015. From the 18,385 participants in the database at the time of analysis, we selected all the participants aged between 18 and 65, without reported history of head trauma or hearing loss, who completed all 3 tests of music perception in the online-test only once and who detected the catch trial inserted in the *Scale* test (Figure 1). The catch trial was a comparison melody in which an obvious
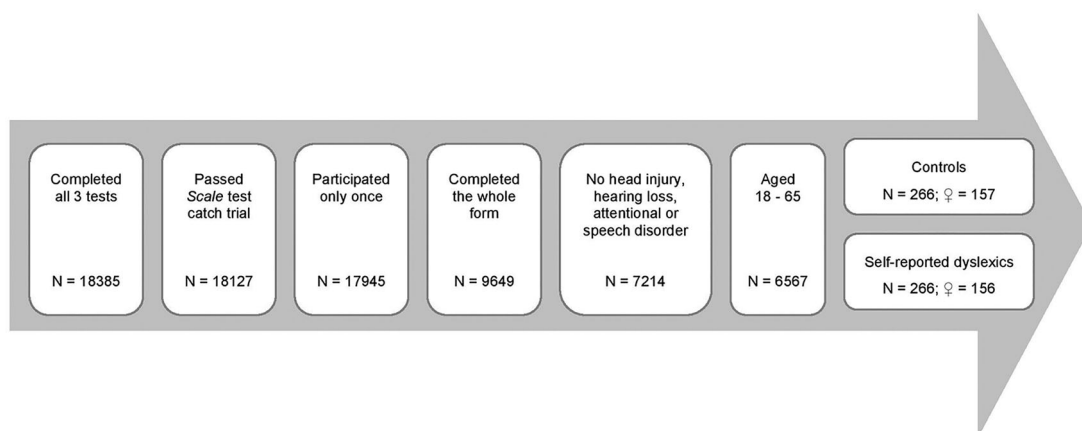
Figure 1. Study 1. Flow-chart for the selection of participants from the on-line database.

pitch change was embedded. Participants who self-reported speech and/or attentional disorders were removed from further analysis in order to exclude cases of ADHD and/or specific language impairments (frequently associated with dyslexia) and to minimize confounding factors.

By these criteria, 6567 participants were considered. Among them, 266 (4.1%) reported to have dyslexia while 6301 did not. We used propensity score matching with a nearest neighbour algorithm (Ho, Imai, King, & Stuart, 2006) to pair self-reported dyslexic participants with a set of controls, taken from the remaining 6301 participants, and matched in age, sex, educational level and musical expertise. The final dataset contained 266 pairs of self-reported dyslexic and control participants (mean age: 32.6 ± 12.5 SD for self-reported dyslexic partipants, 32.8 ± 13.1 for controls; mean years of school education: 17.5 ± 4.0 SD for self-reported dyslexic partipants, 17.1 ± 3.3 for controls; mean years of musical education: 5.9 ± 7.1 SD for self-reported dyslexic partipants, 5.6 ± 7.2 for controls).

Material and tasks

The entire assessment took place online. Participants completed three musical tests – the Scale test, the Off-beat and the Off-key tests – described in detail in Peretz & Vuvan, 2017. The Scale condition comprises 30 pairs of melodies to be judged as same or different. In half of the pairs, the pitch of one note is modified to be out of scale while maintaining the original melodic contour. In the second and third conditions, participants are presented with 24 single melodies and have to detect whether an incongruity occurred. In half of the melodies, a tone is altered either in time (Off-beat condition) or in pitch (Off-key condition). The three musical tests were followed by a self-report inventory collecting biographic data about participant's educational and professional background (4 items), everyday musical habits (9 items), musical ear (10 items), childhood musical experience (8 items), musical education (10 items) and history of various disorders (8 items). Three statements have been identified as key self-descriptions for congenital amusia diagnosis (Peretz et al., 2008; Vuvan et al., 2017): (1) I cannot recognize familiar tunes without the help of the lyrics, (2) I cannot tell if I sing in tune, and (3) I have been told that I sing out-of-tune.

Table 1. Study 1. Mean percentage of correct responses (SD) in the Scale, Off-beat and Off-key tests of the online test, as a function of group.

	Self-reported dyslexics (N = 266)	Controls (N = 266)	T-test		Effect size Cohen's d
			t	p	
Scale	86.7 (9.8)	88.5 (10.3)	2.025	.043	.18
Off-beat	83.7 (10.1)	84.6 (9.6)	1.036	.301	.09
Off-key	83.1 (12.6)	85.9 (11.9)	2.631	.009	.23

Results

Self-report inventory

We analysed the answers relative to participants' everyday musical habits and musical abilities and applied a Bonferroni correction for the nineteen measures considered, setting the alpha threshold to .003. Dyslexic participant's reports regarding their musical habits and musical appreciation attitudes (9 items) did not significantly differ from controls. Nevertheless, in half of the questions related to their musical skills (5/10 items), dyslexic participants rated their abilities significantly more negatively than controls. For example, to the question: "Do you think that you lack a sense of music?" 32.7% responded positively against 20.3% of controls ($\chi^2 = 10.509$, $p = .001$). Over half declared to sing out of tune (56.4% of dyslexic participants vs. 41.0% of controls, $\chi^2 = 12.648$, $p < .001$) and 18.4% declared to be unable to detect when someone does (vs. 9.0% of controls, $\chi^2 = 9.923$, $p = .002$). In addition, a higher proportion of dyslexic participants reported to be unable to follow the beat (24.1% vs. 12.0% of controls, $\chi^2 = 13.015$, $p = .001$). It should be noted, however, that the proportion of individuals who reported to encounter difficulties in familiar tune recognition was quite low but slightly higher among dyslexics (4.9% of dyslexic participants vs. 3.0% of controls, $\chi^2 = 11.089$, $p = .026$).

Table 2. Study 1. Percentage of self-reported dyslexic and control participants performing 2, 1.5 and 1 SD below the mean of controls in Scale, Off-beat and Off-key tests.

	Cut-off	Cut-off score	Self-reported dyslexics (N = 266)	Controls (N = 266)	Chi-square χ^2	p
Scale	−2 SD	67.9	5.3	6.0	.141	.707
	−1.5 SD	73.1	13.9	10.2	1.776	.183
	−1 SD	78.2	19.9	12.03	6.175	.013
Off-beat	−2 SD	65.4	5.6	3.8	1.049	.306
	−1.5 SD	70.2	9.4	4.9	4.081	0.043
	−1 SD	75.0	22.9	18.8	1.378	.24
Off-key	−2 SD	62.2	6.8	5.3	.532	.466
	−1.5 SD	68.1	16.2	9.8	4.813	.028
	−1 SD	74.1	19.5	12.4	5.055	.025

Music perception

Table 1 summarizes the results of the three musical tasks performed by the two groups of participants. For group differences, effects sizes are indicated (Cohen's *d*). Participants with dyslexia scored slightly but significantly lower than controls in *Scale* and *Off-key* conditions but not in *Off-beat*.

Prevalence of amusia

Because the dyslexic and control groups were matched in all respects but self-report of dyslexia, we compared the prevalence of low musical scores between the two groups. For each test, we considered three different thresholds to assess the presence of a deficit: the standard clinical criterion of two standard deviations (−2 SD) below the mean of the matched controls, and 2 more liberal criteria (−1.5 and −1 SD). As can be seen in Table 2, although participants were too few below the stringent −2 SD threshold to yield reliable differences, there was a larger proportion of dyslexics than controls scoring below −1 SD on the *Scale* and *Off-key* tests. Furthermore, there was also a larger proportion of dyslexics than controls scoring below −1.5 SD on the *Off-beat* and *Off-key* tests.

Family history of amusia

According to their report, 18.5% of dyslexic participants had first-degree relatives with a history of musical impairments (singing out-of-tune, cannot recognize familiar songs, no sense of rhythm, do not appreciate or do not enjoy listening to music). The corresponding value was 16.5% in controls, which was not significantly different ($\chi^2 = .003$, $p = .956$).

In sum, we found evidence of slightly lower performance on musical tests in dyslexia compared to matched controls. We also found weak evidence for comorbidity between self-declared dyslexia on the one hand, and low scores (−1 and −1.5 SD) on musical tests. Although these observations are based on a large population, they are limited by the low precision of the literacy measures: self-declared rather than objectively tested and diagnosed dyslexia.

Study 2: Lab testing of adults with diagnosed dyslexia and amusia

Methods

Participants

Data were acquired at two collaborating sites, in Paris and Montreal. Twenty participants with dyslexia (10 females, 10 males) and twenty matched control participants (11 females, 9 males) were tested in Paris. They were recruited through advertisements in Parisian universities. Sixteen participants with congenital amusia (12 females, 4 males) and twenty matched control participants (14 female, 6 males) were tested in Montreal. These Canadian participants were in the local database and had been tested in prior studies. All were paid for their participation.

Inclusion criteria required participants (1) to be a native speaker of French, (2) to report no known neurological/psychiatric disorder, (3) to have no hearing impairment, and (4) to have a non-verbal IQ above 80; (5a) for control participants: to report no known history of reading/oral language difficulties, to score above the 10th centile of ECLA16+ senior high school norms on Pollueur and Alouette standardized reading tests (Gola-Asmussen, Lequette, Pouget, Rouyer, & Zorman, 2010; Lefavrais, 1967), and to be normal at the Montreal Battery of Evaluation of Amusia (MBEA) (Peretz, Champod, & Hyde, 2003). Two Canadian control candidates did not meet inclusion criteria because of deviant reading speeds; they were excluded from further analysis; (5b) for dyslexic participants: self-identification as a dyslexic person and a reading fluency score below the 10th centile of ECLA16+ senior high school norms on Pollueur or Alouette tests (Gola-Asmussen et al., 2010; Lefavrais, 1967), (5c) for amusic participants: a global composite score or a melodic composite score in the

Table 3. Study 2. Mean (SD) age, gender, school education, musical education, non-verbal IQ and verbal IQ as a function of group.

	French dyslexics ($n = 20$)	French controls ($n = 20$)	Canadian amusics ($n = 16$)	Canadian controls ($n = 18$)
Age (years)	28.9 (6.3)	26.3 (4.6)	59.6 (18.4)	60.4 (16.1)
Gender	10 F, 10 M	11 F, 9 M	12 F, 4 M	12 F, 6 M
School education (years)	19.1 (3.2)	19.7 (2.5)	17.4 (2.1)*	15.0 (3.2)
Musical education (years)	2.4 (3.2)	2.7 (3.1)	0.8 (1.2)	1.6 (1.5)
Non-verbal IQ	109.85 (12.23)	108.60 (14.56)	106.94 (16.51)	112.22 (11.91)
Verbal IQ	110.05 (15.06)	119.05 (13.75)	112.19 (11.74)	113.22 (6.38)

*Score is significantly different from controls at $p < .05$ level.

MBEA two standard deviations below the average of the general population (Peretz et al., 2003).

Amusic and dyslexic participants were matched to controls in terms of nationality, age, musical education and non-verbal intelligence, as assessed by the Matrix and Picture Completion tasks of the Wechsler Adult Intelligence Scale III (WAIS-III, Wechsler, 2000) (Table 3). Their verbal intelligence was assessed by using the Vocabulary and Similarities subtests of the Wechsler Adult Intelligence Scale (French version of the WAIS-III; Wechsler, 2000). There was a significant difference in school education between Canadian amusic participants and Canadian controls (mean years: 17.4 ± 2.1 SD for Canadian amusic participants vs. 15.0 ± 3.2 SD for Canadian controls, two-tailed t-test: t (32) = 2.602, p = .014). However, this variable did not correlate with any dependent variable (i.e., literacy, phonology and music perception measures). French and Canadian participants significantly differed in age (mean years: 27.6 ± 5.6 SD for French participants vs. 60.0 ± 16.9 SD for Canadian participants, two tailed t-test: t (72) = −11.405, $p <$ .001), school education (mean years: 19.4 ± 2.8 SD for French participants vs. 16.2 ± 3.0 SD for Canadian participants, two-tailed t-test: t (72) = 4.787, $p <$ .001) and music education (mean years: 2.6 ± 3.1 SD for French participants vs. 1.2 ± 1.4 SD for Canadian participants, two-tailed t-test: t (72) = 2.329, $p <$ .023) but not in non-verbal IQ (mean: 109.23 ± 13.29 SD for French participants vs. 113.56 ± 14.09 SD for Canadian participants, two-tailed t-test: t (72) = −1.360, p = 178) nor verbal IQ (mean: 114.55 ± 14.94 SD for French participants vs. 112.74 ± 9.16 SD for Canadian participants, two-tailed t-test: t (72) = .614, p = .541).

Participants filled out the French version of the Adult Reading History Questionnaire (Lefly & Pennington, 2000), a 23-item self-report screening tool designed to measure risk of reading disability in adults. Each item of the questionnaire is responded to with a 5-point Likert-type scale ranging from 0 to 4, resulting in a score range of 0–92. The total score is divided by the maximum possible score (92) to generate a percentage score ranging from 0 to 1, such that higher scores represent greater reading difficulties. A threshold of 0.40 is a reliable predictor of a diagnosis of reading disability (Lefly & Pennington, 2000). French dyslexic participants scored significantly higher than French controls and all of them reported a history of reading difficulties (mean

scores/1: $0.58 \pm .14$ SD for French dyslexic participants vs. $0.19 \pm .28$ SD for French controls, two-tailed t-test: t (38) = 5.467, $p <$ .001). There was no significant difference between Canadian amusic participants and Canadian controls in the mean score of this questionnaire (mean scores/1: 0.27 ± 0.18 SD for Canadian amusic participants vs. $0.25 \pm .06$ SD for Canadian controls, two-tailed t-test: t (32) = .538, p = .59). A majority of Canadian participants with amusia reported a high interest for reading (11/16) and to have experienced no difficulty in learning to read during childhood (11/16); most of them judged their actual reading speed (15/16) and orthographic abilities (15/16) average or above average. However, Canadian amusic participants reported significantly more first-degree relatives with reading difficulties than Canadian controls (12.4% for Canadian amusic participants vs. 3.0% for Canadian controls, χ^2 = 6.014, p = .014).

In the self-assessment part of the On-line Identification Test of Congenital Amusia (Peretz et al., 2008), a majority of Canadian amusic participants (81.3%) declared to lack a sense of music and their responses to the three key statements for congenital amusia diagnosis (see above) do discriminate them from Canadian controls' responses (81.3% of Canadian amusic participants vs. 5.6% of Canadian controls responded positively to at least one of the three statements, χ^2 = 20.896, $p <$ .001). French dyslexic participant's reports did not differ from French controls regarding their musical habits and musical appreciation attitudes. A majority (75.0%) reported to listen often or very often to music intentionally, and listening to music was "a pleasure" for all of them. Yet, 50% of the French dyslexic participants vs. 15% of the French controls responded positively to at least one of the three statements that are diagnostic of congenital amusia (χ^2 = 4.103, p = .043). Moreover, French dyslexic participants generally rated their abilities more negatively than French controls. Futhermore, French dyslexic participants reported significantly more first-degree relatives with musical difficulties than French controls (22.6% for French dyslexic participants vs. 6.8% for French controls, χ^2 = 10.631, p = .001).

Ethical approval was granted by the local Ethics Committees (CPP Bicêtre in France; University of Montreal in Canada) and all participants gave written informed consent.

Experimental battery

Music perception and memory. Music perception and memory was assessed using the Montreal Battery of Evaluation of Amusia (MBEA) (Peretz et al., 2003), which involves six tests measuring scale, contour, interval, rhythm, metre, and memory of unfamiliar but conventional melodies. Each test comprises 30 trials and uses the same pool of 30 unfamiliar melodies that are written according to the rules of the Western system. The *Scale, Contour*, *Interval*, and *Rhythm* tests involve pairs of melodies and consist of a same-different judgement. The *Metric* task requires participants to categorize harmonized melodies as being marches (duple metre) or waltzes (triple metre), and the *Memory* task, to recognize a melody as having been presented earlier during the session or not. Amusia is diagnosed when an individual performs two standard deviations below the mean performance of the general population in the global composite score (mean score of the 6 tests) or in the melodic composite score (mean score of the *Scale*, *Contour*, and *Interval* tests) without failing any catch trials.

Literacy and phonological skills. *Reading and orthographic skills.* Reading skills were assessed by the standardized French reading test "L'alouette" (Lefavrais, 1967). This meaningless text comprises 265 words ranging from common to rarely used words. Participants are instructed to read outloud the text as fast and as accurately as possible. Reading fluency scores are computed by combining total reading time and reading errors. In addition, participants completed the *Text reading*, *Word reading* (regular, irregular, pseudowords), *Text dictation*, and *Word dictation* (regular, irregular, pseudowords) test of the French ECLA-16+ standardized battery (Gola-Asmussen et al., 2010). Both accuracy and speed were scored.

Phonological working memory. (i) *Digit span.* Forward and backward digit span (from the French version of the WAIS-III; Wechsler, 2000) were used to compute age-appropriate scaled scores in order to obtain a measure of phonological working memory. (ii) *Pseudoword repetition.* Participants were verbally presented with pseudowords of 5 and 7 syllables and were instructed to repeat them as accurately as possible.

Phonological awareness. (i) *Initial Phoneme Deletion.* Participants were required to delete the first sound of verbally presented words and to pronounce the remaining word. Both accuracy and speed were scored. (ii) *Spoonerisms.* Participants were verbally presented with pairs of words and were instructed to swap the first sound of the two words and then to pronounce the resulting pseudowords while maintaining their correct order. A composite score taking into account both accuracy and speed was computed.

Rapid Automatized Naming (RAN). Participants named series of 50 items (objects, colours or digits) as fast as possible. Each naming test was administered twice with different sheets. The score was the sum of total naming time for both sheets of each test.

Results

Music perception and memory

Performance of amusic, dyslexic and matched control participants was assessed using signal detection theory (Macmillan & Creelman, 2004). We calculated d' sensitivity index and criterion c as $d' = z(H) - z(FA)$ and $c = -0.5 * [z(H) + z(FA)]$. Hits and false alarms were defined as follows: for *Scale*, *Contour*, *Interval*, and *Rhythm* tests: $H = p$ (response = different | stimulus = different) and $FA = p$ (response = different | stimulus = same), for *Metric*: $H = p$ (response = waltz | stimulus = waltz) and $FA = p$ (response = waltz | stimulus = march), and for *Memory:* $H = p$ (response = old (stimulus = old) and $FA = p$ (response = old | stimulus = new).

As expected, Canadian amusic participants scored significantly lower than Canadian controls in the MBEA (Table 4). Their low scores were due to a high rate of misses (mean: 55.6% in the melody discrimination tests): they reported most of the time hearing identical melodies, which is in line with the fact that amusic individuals do not consciously perceive pitch violations (e.g., Peretz et al., 2009). Accordingly, the amusic group had significantly higher decision criteria than controls (mean = .40 ± .41 SD for Canadian amusic individuals vs. mean = −.03 ± .19 SD for Canadian controls, two-tailed *t*-test: *t* (31) = −3.869, *p* = .001).

French dyslexic participants did not significantly differ from French controls in the MBEA (Table 4). Decision criteria did not significantly differ between groups (mean = .01 ± .59 SD for French dyslexic individuals vs. mean = −.11 ± .33 SD for French controls, two-tailed *t*-test: *t* (38) = −.770, *p* = .446) and were on

Table 4. Study 2. MBEA, Literacy and phonology mean z-scores (SD) as a function of group.

	French dyslexics ($n = 20$)	French controls ($n = 20$)	Canadian amusics ($n = 16$)	Canadian controls ($n = 18$)
MBEA global z-score	−1.01 (1.98)	.03 (1.30)	−4.43 (1.82)***	0.00 (1.00)
Literacy factor	−5.67 (3.78)***	0.00 (1.00)	−1.16 (1.68)	−0.46 (0.70)
Reading	−5.21 (3.56)***	0.00 (1.00)	−1.87 (1.60)	−1.34 (0.77)
Orthography	−3.81 (2.91)***	0.00 (1.00)	0.02 (1.17)	0.60 (0.62)
Phonology factor	−1.92 (1.25)***	0.00 (1.00)	−0.80 (1.37)	−0.10 (0.88)
Phonological working memory	−1.28 (1.10)***	0.00 (1.00)	−0.29 (1.15)	−0.17 (0.71)
Phonological awareness	−0.83 (0.69)**	0.00 (1.00)	−0.45 (0.94)	0.28 (1.26)
Rapid Automatized Naming	−2.12 (1.88)***	0.00 (1.00)	−1.06 (1.57)	0.33 (0.92)

Notes: A Reading score was calculated from "L'alouette", *Text Reading* and *Word Reading* scores; an Orthographic score from Word *Dictation* and *Text Dictation* scores. A Phonological Working Memory score was calculated from *Digit Span* and *Pseudo Word* Repetition scores; a Phonological Awareness score from *Initial Phoneme Deletion and Spoonerism* scores; a Rapid Automatized Naming score as the mean score of picture, digit and colour naming subtests. *Z*-scores were computed from the means and standard deviations of the Canadian controls for the MBEA; they were computed from the means and standard deviations of the French controls for literacy and phonological measures.
**Score is significantly different from controls at $p < .01$ level.
***Score is significantly different from controls at $p < .001$ level.

average not significantly different from zero (mean = −0.05 ± 0.48 SD, one-sample *t*-test: t (39) = −.632, $p = .531$), indicating no response bias.

However, six French dyslexic participants out of twenty (30%) performed under the criterion for amusia diagnosis (two standard deviations below the mean of the general population, cut-off: 21.9/30 on global score and 22.1/30 on melodic composite score (Peretz et al., 2003; Figure 2).

Because control participants were selected in such a way as to exclude amusia, we refered to population-based prevalence rates. Such a proportion of 6 out of 20 tends to be significanty higher from what would be expected from the population prevalence of amusia (0.3 out of 20 expected according to the conservative value of 1.5%, Yates' $\chi^2 = 4.162$, $p = .041$; 0.8 out of 20 expected according to the common value of 4%, Yates' $\chi^2 = 3.125$, $p = .077$). These 6
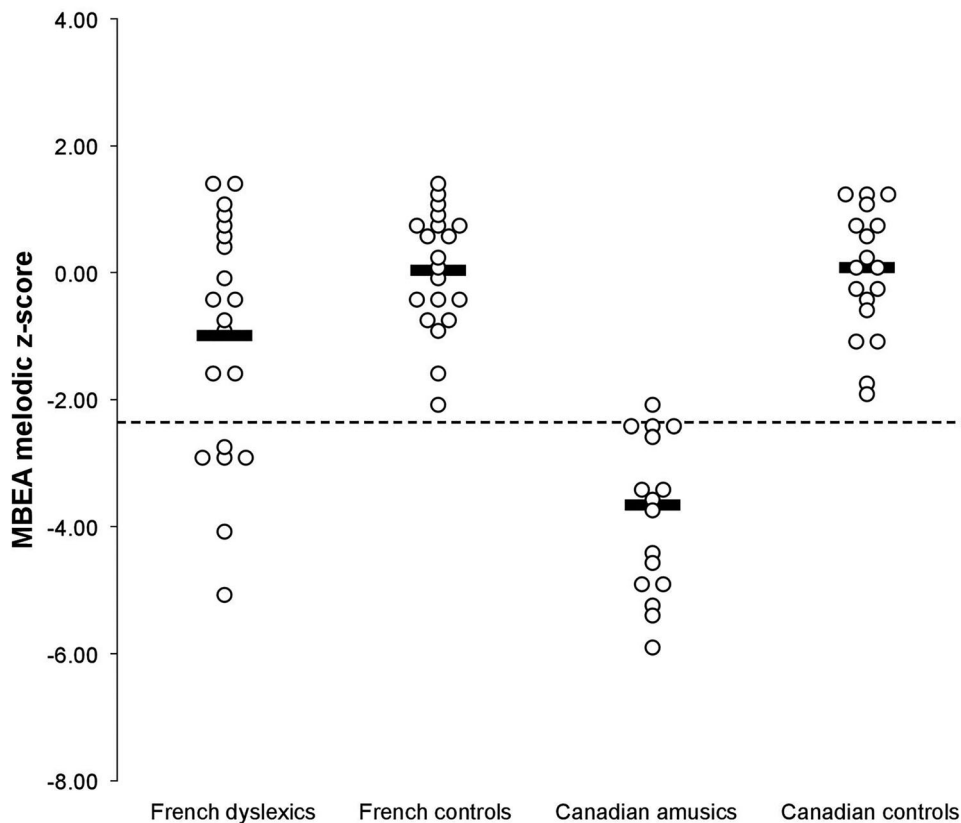
Figure 2. Study 2. Distribution of dyslexic, amusic and control participants along their melodic composite *z*-scores (computed from the mean and standard deviation of the Canadian controls) in the MBEA. Black lines indicate the group median. Six French dyslexic participants performed under the cut-off (dashed line) for amusia diagnosis. The Canadian amusic individual situated above the cut-off had a global composite score two standard deviations below the mean performance of the general population.
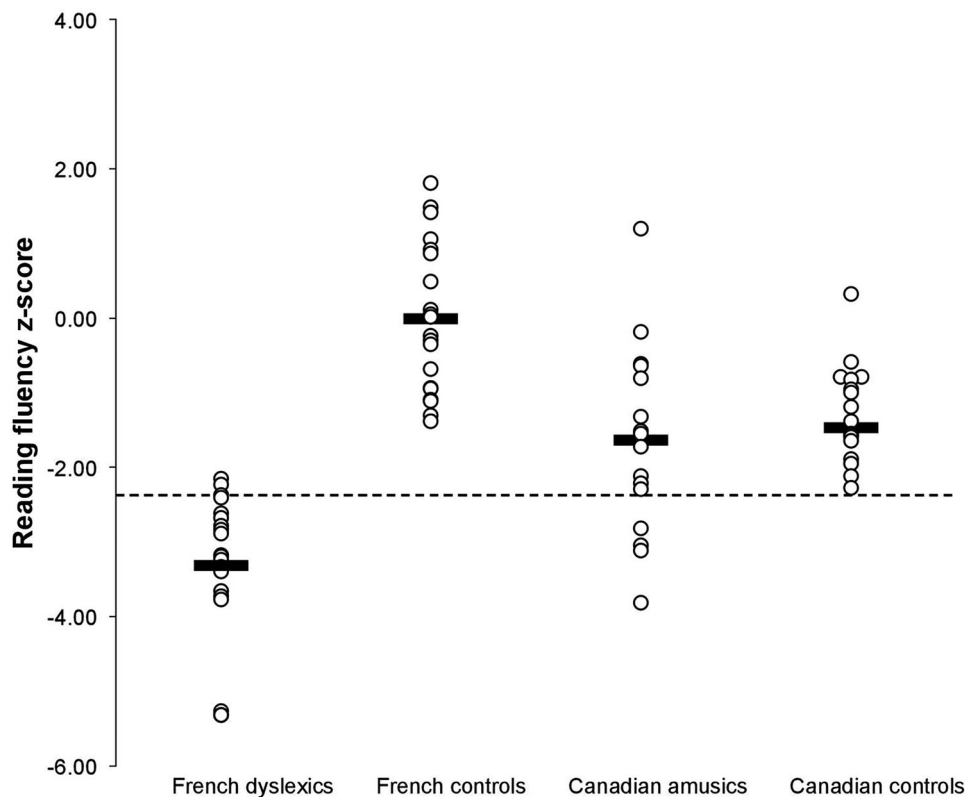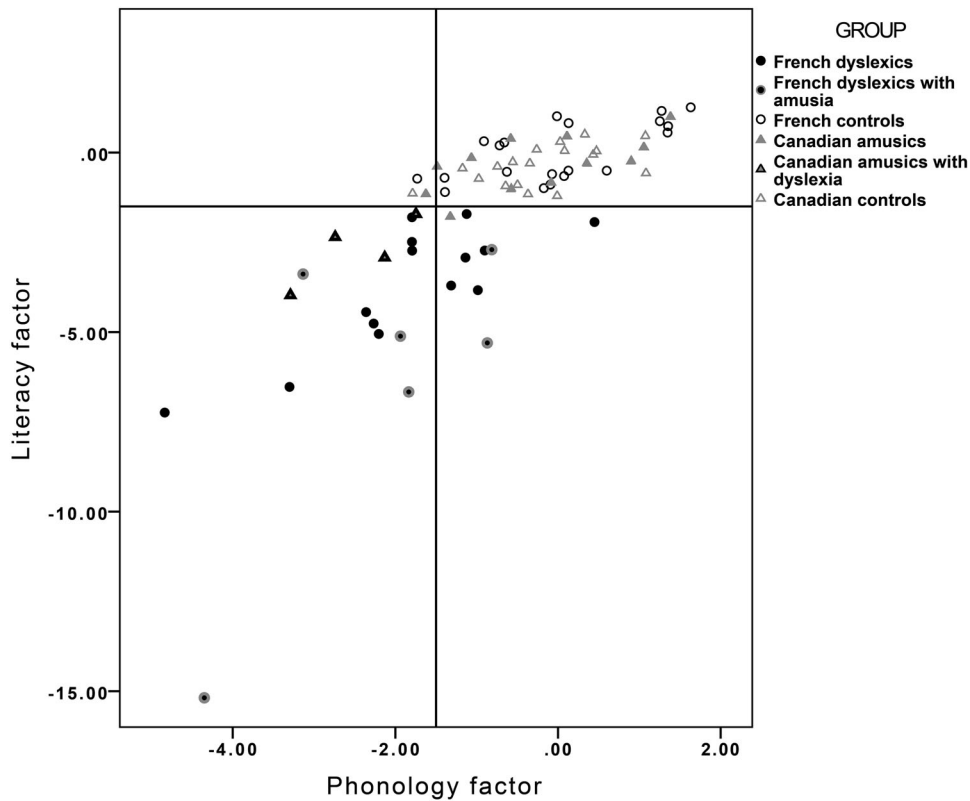
Figure 3. Study 2. Distribution of dyslexic, amusic and control participants along their z-scores (computed from the mean and standard deviation of the French controls) in the standardized reading test "L'alouette". Black lines indicate the group median. Four Canadian amusic participants performed under the cut-off (dashed line) for dyslexia diagnosis. The two French dyslexic individuals situated above the cut-off performed below the 10th centile of ECLA16+ senior high school norms on Pollueur test.

French dyslexic individuals with amusia differed significantly from French controls in each melodic test (*Scale*: mean d': 1.99 ± 1.11 SD for French dyslexic individuals with amusia vs. 4.54 ± 1.42 SD for French controls, Mann–Whitney U-test: U = 8.000, p = .001, *Contour*: mean d': 1.50 ± 1.00 SD for French dyslexic individuals with amusia vs. 3.90 ± 1.78 SD for French controls, U = 10.000, p = .001, *Interval*: mean d': 1.01 ± 1.12 SD for French dyslexic individuals with amusia vs. 3.50 ± 1.73 SD for French controls, U = 12.000, p = .002) and in *Memory* (mean d': 2.26 ± 0.96 SD for French dyslexic individuals with amusia vs. 4.99 ± 1.86 SD for French controls, U = 7.000, p < .001). They also differed, to a smaller extent, in *Rhythm* (mean d': 3.11 ± 1.19 SD for French dyslexic individuals with amusia vs. 4.97 ± 2.16 SD for French controls, U = 24.500, p = .028) but not in *Metric*: mean d': 2.01 ± 1.50 SD for French dyslexic individuals with amusia vs. 3.35 ± 2.74 SD for French controls, U = 49.000, p = .533). Their poor performances in the melodic tests were also due to a high rate of misses (mean: 53.7%). In line with these findings, each French

dyslexic participant with amusia reported musical difficulties related with pitch processing abilities in the self-assessment part of questionnaire.

Literacy and phonological skills
Literacy. As expected, French dyslexic participants scored significantly lower than French controls in literacy measures (Table 4). At the group level, Canadian amusic participants and Canadian controls did not differ much in their literacy performance. No significant difference in reading and orthographic scores was observed between groups (Table 4). Nonetheless, at an individual level, four Canadian amusic participants out of sixteen (25%) performed under the criterion for dyslexia (reading fluency score below the 10th centile of ECLA16+ senior high school norms on Pollueur or Alouette tests, cut-off: 153 on Pollueur and 127 on Alouette (Gola-Asmussen et al., 2010); Figure 3). Three of them also exhibited an Adult Reading History Questionnaire score consistent with reading disability (mean score >0.40, [Lefly & Pennington, 2000]). Again, as control participants were

Figure 4. Study 2. Distribution of individual composite z-scores along the literacy and phonology factors. The horizontal and vertical lines indicate the −1.5 standard deviation threshold.

selected in such a way as to exclude dyslexia, we refered to population-based prevalence rates. This proportion of 4 out of 16 did not significantly differ from the population prevalence of dyslexia (0.48 out of 16 expected according to the minimal value of 3%, Yates' $\chi^2 = 1.648$, $p = .199$; 1.12 out of 16 expected according to the maximal value of 7%, Yates' $\chi^2 = .822$, $p = .365$).

Phonological skills. As can be seen in Table 4, French dyslexic participants scored significantly lower than French controls in all phonology measures, which attests to the presence of a phonological deficit. There was no significant difference between Canadian amusic participants and Canadian controls in phonological working memory, phonological awareness, nor rapid automatized naming tasks.

Correlations. We took as the literacy factor the average of reading and orthography z-scores, and as the phonology factor the average of phonological working memory, phonological awareness and Rapid Automatized Naming z-scores. Figure 4 shows

participants' individual scores on the two factors. As expected from a large body of research on literacy, we observed a positive correlation between phonological and literacy skills, across all groups and also within each group except Canadian controls (All: r (70) = .753, $p < .001$; French dyslexic participants: r (16) = .496, $p = .036$; French controls: r (16) = .741, $p < .001$; Canadian amusic participants: r (12) = .810, $p < .001$; Canadian controls: r (14) = .454, $p = .078$, after partialling out age and nonverbal IQ). The four Canadian amusic individuals with dyslexia were impaired in both literacy and phonological skills, as would be expected from typical dyslexic cases.

Joint analysis of music perception and literacy measures

We observed positive correlations across groups between musical and literacy skills as well as between musical and phonological skills, as represented by our composite variables (r (70) = .246, $p = .037$; r (70) = .267, $p = .024$, after partialling out age and nonverbal IQ). However, these correlations
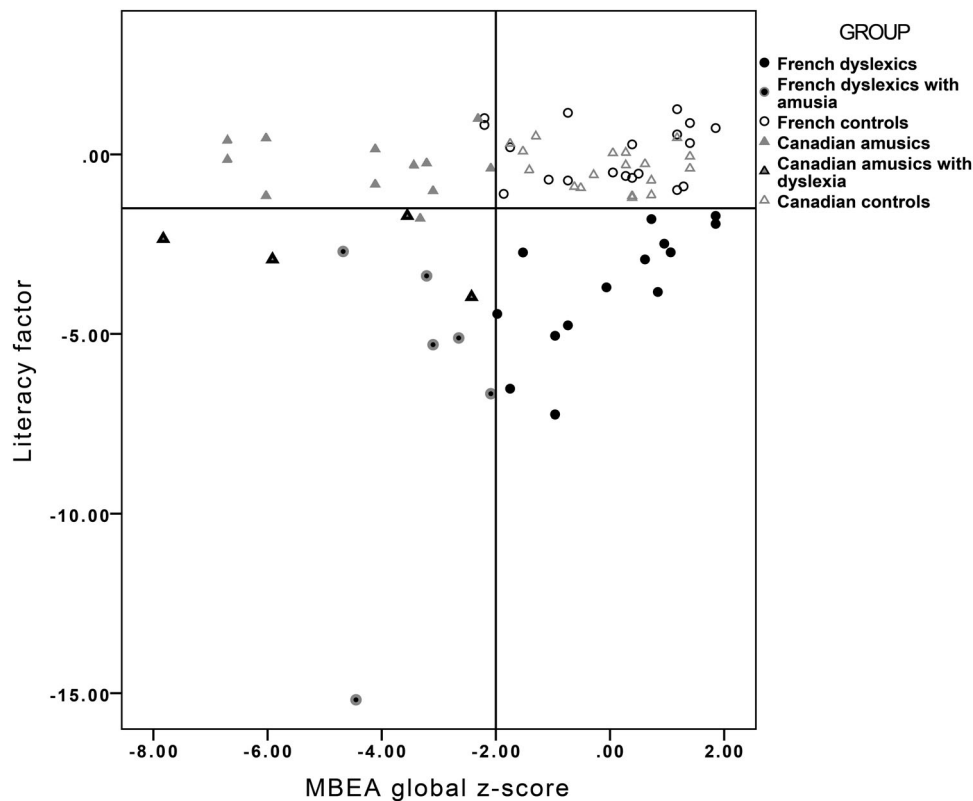
Figure 5. Study 2. Distribution of individual composite *z*-scores along the literacy and musical (MBEA global) factors. The horizontal and vertical lines indicate the −1.5 and −2.0 standard deviation threshold.

did not reach significance in each group (French dyslexic participants: r (16) = .373, p = .127; r (16) = .133, p = .600; French controls: r (16) = .059, p = .817; r (16) = .393, p = .107; Canadian amusic participants: r (12) = .024, p = .934; r (12) = .226, p = .438; Canadian controls: r (14) = −.417, p = .108; r (14) = −.191, p = .479). As can be seen in Figure 5, there is no general relationship between musical and literacy skills, unlike what is observed between phonological and literacy skills.

General discussion

The present study aimed to investigate at a behavioural level whether there is comorbidity and cognitive overlap between developmental dyslexia and congenital amusia in adults. For this purpose, we analysed the database of a large population of volunteers taking an online test of musical perception, with a focus on individuals reporting dyslexia compared to those not reporting dyslexia (Study 1); In a distinct study, we administered a whole battery of literacy and musical tests to two groups of participants

diagnosed with dyslexia and with amusia respectively (Study 2). Overall the two studies have different advantages and limitations, and converge on the suggestion of a limited but significant comorbidity (25%–30%) between dyslexia and amusia.

In study 1, consistent with previous studies on musical abilities in dyslexia (Santos et al., 2007; Ziegler et al., 2012), we found that participants who self-reported dyslexia also reported more musical difficulties than other matched participants, and scored slightly but significantly lower in the two pitch-related conditions. Furthermore, self-reported dyslexic individuals were overrepresented amongst low-scorers at the musical tests (about 20% below −1 SD compared to 12% of controls); applying a more stringent deviance thresholds (−2 SD) yielded too few individuals to address comorbidity with amusia.

In contrast with prior reports of temporal processing deficits in dyslexia (Flaugnacco et al., 2014; Goswami, 2011; Goswami, Huss, Mead, Fosker, & Verney, 2013; Huss et al., 2011), participants with self-reported dyslexia and controls did not differ in rhythm perception performance. Using a comparable

initial sample, Peretz and Vuvan (Peretz & Vuvan, 2017) found that a deficit in the *Off-beat* test seemed associated with many other neurodevelopmental disorders including dyslexia. Yet, the approach used here was slightly different, with a focus on individuals reporting dyslexia. In addition, we used more stringent inclusion criteria for dyslexia in terms of exclusion of comorbidities (attentional and speech disorders). Thus, the association of rhythm perception deficits with dyslexia may have more to do with other confounding factors than with dyslexia per se.

This first study had the advantage of considering a very large sample of participants. The main limitation, though, is that the inclusion as dyslexic or control participant was based on self-report: there was no objective measure of literacy performance and we have no assurance that participants had a proper understanding of dyslexia. This might induce various biases, in particular depending on participants' age, dyslexia being nowadays more systematically detected by the school system. Dyslexia is also a "popular diagnosis" that is often used in common language instead of other diagnostic categories. The self-reported dyslexia in this study may therefore lack specificity. To counteract this trend, we excluded individuals with self-reported speech or attention problems. In addition, between-group differences in the report of musical difficulties might be due to over-reporting bias: individuals reporting dyslexia might tend to report more deficits in general. These subjective reports are nevertheless consistent with the objective measures obtained in the musical tests. Another limitation of this study is that, as discussed in a previous article (Vuvan et al., 2017), the On-line Test is a screening tool for congenital amusia, but is not sufficient to establish a diagnosis. Thus, the proportion of amusic individuals in the sample may have been underestimated. Finally, potential confounding factors, such as general intelligence, are not controlled for. With all these considerations in mind, Study 1 does provide a first hint that at least some individuals with dyslexia may score more poorly on musical tests.

Study 2 addressed the same question from a different and complementary vantage point, using rich data and rigorous diagnostic criteria. At the group level, French dyslexic participants did not significantly differ from French controls in any subtest of the MBEA, which is relatively unsurprising given the small group sizes. Nevertheless, at an individual level, 6 cases out of 20 (30%) French dyslexic participants showed performance in musical tests consistent with a diagnosis of congenital amusia. These 6 individuals performed like controls in the metric test and showed impaired performance in melody and rhythm discrimination, although the effect size for the rhythm test was smaller than that for the three melodic tests. This pattern of preserved performance on temporal tasks and impaired performance in pitch-related discrimination is consistent with the canonical description of congenital amusia (Peretz, 2016), and also with previous research indicating that the mere presence of pitch in temporal tasks may compromise amusics' performance (Phillips-Silver, Toiviainen, Gosselin, & Peretz, 2013).

Turning to literacy, Canadian amusic participants did not differ at the group level from Canadian controls in reading and orthography scores, but at an individual level, 4 cases out of 16 (25%) Canadian amusic participants had reading scores consistent with a diagnosis of dyslexia. It should be noted that Canadian controls scored about one standard deviation below French controls in reading tasks. This could be due to differences in education between the two groups, as well as to the fact that the French texts were less appropriate for Canadian French speakers. Nevertheless, the Canadian control and amusic participants were closely matched in education level, and the four Canadian amusic individuals diagnosed with dyslexia scored two standard deviations below their matched controls in the Alouette reading test, so their comorbid status is not in question, regardless of differences between the two countries. In contrast with previous findings (Jones et al., 2009; Sun et al., 2017), Canadian amusic and Canadian control participants did not significantly differ in phonological measures. However, the Canadian amusic individuals with reading disability also showed poor performance in phonological tests, consistent with a diagnosis of dyslexia. It is therefore possible that previous reports of group differences in phonological skills between amusic and control participants might be due to a subgroup of participants with comorbid amusia and dyslexia.

Finally, in accordance with previous studies reporting links between musical skills and literacy performances (Anvari, Trainor, Woodside, & Levy, 2002; Cogo-Moreira, Brandão de Ávila, Ploubidis, de Jesus Mari, & Najbauer, 2013; Flaugnacco et al., 2014; Loui

et al., 2011) we observed a positive correlation between MBEA scores and literacy measures across the 4 groups of participants. However, these correlations were not significant within each group, and were carried by the few individuals with difficulties in both domains, thus suggesting that this is not a general relationship.

Overall, Study 2 presents the opposite advantages and limitations from Study 1: a large test battery covering both areas was used, identical in both populations, but in a small sample of participants which does not allow a sufficiently reliable estimation of the prevalence. Taken together, both studies are consistent with a moderate comorbidity between dyslexia and amusia. This observation is strengthened by the fact that our sample of dyslexic and control participants was high-level achieving (more than 19 years of school education in average) so potentially more resourceful, socially privileged and with more access to remediation, thus decreasing the likelihood of comorbidity.

Our results have been obtained in French and may not generalize to all languages. Indeed, it may be that the relationship between dyslexia and amusia varies as a function of the melodic demands of each language and writing system. In particular, it has been suggested that amusia might have consequences on speech processing in tone languages (e.g., Nan et al., 2010). Whether it also has consequences on literacy skills may depend on whether and how tone information is transcribed in the writing system.

Weiss et al. (2014) have drawn attention to the "enigma of dyslexic musicians". Musicians are highly trained to improve auditory working memory, auditory attention, and conscious access to sounds, and seem to have specialized auditory processing skills (Kraus & Chandrasekaran, 2010; Zatorre, Chen, & Penhune, 2007). Yet, they can also have persistent dyslexia (e.g., Bishop-Liebler, Welch, Huss, Thomson, & Goswami, 2014). This is only puzzling if one assumes that dyslexic individuals are also amusic, or that musical abilities are tightly linked with phonological abilities. However, our results suggest that this may simply not be the general case. Indeed, dyslexic and control musicians in Weiss et al. (2014) did not differ in their basic auditory and rhythmic skills. However, dyslexic musicians remained impaired in working memory for melodic and rhythmic patterns, suggesting that (1) a deficit circumscribed to auditory

working memory may not in itself be an impediment to musical performance, and (2) that intensive musical training is insufficient to restore auditory working memory deficits and to remediate reading disability.

A comorbidity between developmental dyslexia and congenital amusia can potentially be accounted for in several different ways and at several levels of explanation. Firstly, the association could be spurious in the sense of reflecting more general developmental anomalies that affect cognitive functioning in general. However, the six cases of dyslexia with amusia and the four cases of amusia with dyslexia did not differ from control participants in general intellectual functioning. Thus, a general developmental vulnerability does not seem a plausible explanation here.

If the association is genuine, it may reflect shared processes at either etiological, neural, or cognitive level. In terms of aetiology, it is interesting to note that in Study 2, Canadian amusic participants reported more relatives with reading problems than controls, and dyslexic participants reported more relatives with music problems than controls, consistent with the idea of shared heritable factors. However, too little is currently known about the genetic basis of amusia to assess whether there are any associated genes shared with dyslexia.

Association may arise at the neural level due to the proximity of brain regions involved in amusia and in dyslexia (in particular, superior temporal et inferior frontal cortex [Peretz, 2016; Ramus, Altarelli, Jednoróg, Zhao, & Scotto di Covella, 2018]). According to one model, a combination of a general susceptibility mechanism (such as neuronal migration disruption) and of additional factors determining the precise location of the disruptions would increase the likelihood that neighbouring brain regions are affected together, even when they are functionally independent, thus inducing comorbidities between different disorders (Ramus, 2004). Another related idea is that amusia and dyslexia might be hemispheric mirror images, involving a disruption of superior temporal-inferior frontal networks, in the left for dyslexia and in the right for amusia (Boets et al., 2013; Peretz, 2016). The underlying mechanisms might be such that a bilateral disruption would arise more often than expected from the prevalence of left and right disruptions.

Finally, comorbidity may arise at the cognitive level, if one or several cognitive deficits are risk factors for both disorders. There are at least two possibilities in this regard. One is that the same basic auditory processing deficit is a risk factor for both dyslexia and amusia. While this hypothesis seems superficially consistent with auditory theories of dyslexia, the specific types of auditory deficits usually postulated in dyslexia (such as rapid temporal processing, e.g., Farmer & Klein, 1995, or amplitude rise time processing, Goswami, 2011) are not the same as the pitch perception deficits postulated in amusia (Tillmann, Albouy, & Caclin, 2015). Thus this hypothesis does not seem supported (but Ziegler et al., 2012 and Albouy et al., 2016 argue otherwise). It could still be the case that the two types of auditory deficits co-occur more often than expected, because of the proximity or homology of the underlying brain regions, as explained above.

Alternatively, shared deficits might be situated at higher levels of processing. As mentioned in the introduction, both amusia and dyslexia can be conceptualized as disorders of conscious access and short-term memory processes applied to sounds (Loui et al., 2011; Peretz et al., 2009; Ramus & Ahissar, 2012; Tillmann et al., 2016). This deficit has been documented in each domain, that is, for musical pitch in the case of amusia and for speech sounds in the case of dyslexia. Deficits of higher-order representation of musical timbre have also been reported in congenital amusia (e.g., Marin, Gingras, & Stewart, 2012; Tillmann, Schulze, & Foxton, 2009), which might be closer to phonological processing. We hypothesise that cases of comorbidity might arise from an access deficit applied to both musical and speech sounds, if such cases occur more often than expected. Our finding of poorer performance in phonological awareness tests in individuals with both amusia and dyslexia is consistent with this idea. So is the finding by Sun et al. (2017) that a significant proportion (40%) of their amusic sample exhibited impairments in phonological awareness. Further research will be needed to determine precisely at which level of description (etiological, neural, cognitive) comorbidity between dyslexia and amusia arises, and if at the cognitive level, exactly which shared deficits are involved.

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