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Comorbidity and cognitive overlap between developmental dyslexia and congenital amusia in children

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A B S T R A C T

Developmental dyslexia and congenital amusia are two specific neurodevelopmental disorders that affect reading and music perception, respectively. Similarities at perceptual, cognitive, and anatomical levels raise the possibility that a common factor is at play in their emergence, albeit in different domains. However, little consideration has been given to what extent they can co-occur. A first adult study suggested a 30% amusia rate in dyslexia and a 25% dyslexia rate in amusia (Couvignou et al., *Cognitive Neuropsychology* 2019). We present newly acquired data from 38 dyslexic and 38 typically developing children. These were assessed with literacy and phonological tests, as well as with three musical tests: the Montreal Battery of Evaluation of Musical Abilities, a pitch and time change detection task, and a singing task. Overall, about 34% of the dyslexic children were musically impaired, a proportion that is significantly higher than both the estimated 1.5–4% prevalence of congenital amusia in the general population and the rate of 5% observed within the control group. They were mostly affected in the pitch dimension, both in terms of perception and production. Correlations and prediction links were found between pitch processing skills and language measures after partialing out confounding factors. These findings are discussed with regard to cognitive and neural explanatory hypotheses of a comorbidity between dyslexia and amusia.

1. Introduction

Developmental dyslexia is characterized as a specific and persistent impairment in the development of reading skills that cannot be accounted for by mental age, inadequate schooling, or obvious sensory or neurological damage (World Health Organization, 2011). This common learning disorder, which affects about 3–7% of schooled children (Lindgren et al., 1985), can be severely invalidating, being a risk factor for increased anxiety, depression and academic failure (Boetsch et al., 1996).

Over the last several decades, a large body of evidence have supported phonological impairments as the main underlying cognitive cause of the reading disabilities in dyslexia (Ramus, 2003; Vellutino et al., 2004). This so-called *phonological deficit* is characterized by low phonological awareness, reduced phonological memory and slow lexical retrieval (Snowling, 2000; Wagner and Torgesen, 1987) and seems to play a causal role in the development of poor reading skills, as shown by longitudinal studies of children at familial risk for dyslexia (Lyytinen et al., 2004; Puolakanaho et al., 2007). Nevertheless, there is still debate as to whether or not more primary auditory deficits underlie the phonological deficit (Goswami, 2015; Hornickel and Kraus, 2013), as well as about the specific nature of the phonological deficit (Boada and Pennington, 2006; Ramus and Szenkovits, 2008). Visual or visual-attentional deficits have been proposed as an alternative proximal cause of dyslexia (Stein and Walsh, 1997; Valdois et al., 2004; Vidyasagar and Pammer, 2010), but may affect only a subset of individuals (Ramus et al., 2003; Saksida et al., 2016).

A remarkable feature of developmental dyslexia is that it rarely occurs alone. Indeed, dyslexia is frequently associated with a constellation of other learning disabilities such as developmental language disorder (up to 50%), motor disorders such as dyspraxia, coordination disorders or dysgraphia (up to 50%), dyscalculia (about 40%) and attention deficit disorders (about 30%; Germanò et al., 2010; Kaplan et al., 1998; McArthur et al., 2000; Wilson et al., 2015). Overall, there are many

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comorbidities among neurodevelopmental disorders, which tend to aggregate with each other. It has been proposed that they share a common biological origin, which may reside in abnormal neuronal migration (Galaburda et al., 1985; Galaburda and Kemper, 1979; Ramus, 2004).

In addition, a handful of studies have reported musical impairments in dyslexia. These difficulties are diverse in nature, sometimes concerning perception, sometimes production of rhythm, pitch or both (Baldeweg et al., 1999; Lifshitz-Ben-Basat and Fostick, 2019; Overy et al., 2003; Thomson and Goswami, 2008; Ziegler et al., 2012). Besides, it is not always clear whether there are actual group differences or whether these are rather driven by a small number of individuals with particularly low performance. Specifically, little consideration has been paid to the extent to which dyslexia may coexist with congenital amusia.

Congenital amusia is a lifelong musical disorder that cannot be explained by hearing loss, brain injury, intellectual disability or lack of exposure to music. Its most common (or most studied) form is characterized by an inability to recognize a familiar melody without the aid of lyrics, to detect false notes, and to judge whether someone is singing out of tune (Peretz, 2013). Some amusic individuals also report not enjoying music. The main explanatory cognitive hypothesis is that of a deficit in pitch processing. Seminal studies have focused on impairments of fine-grained pitch discrimination, but more recent findings point to deficits in pitch short-term memory (for a review, see Tillmann et al., 2016b). Congenital amusia has been estimated to affect 1.5–4% of the population (Peretz and Vuvan, 2017) but only a few cases were reported in childhood (Lebrun et al., 2012; Mignault Goulet et al., 2012). Thus, although the hypothesis of a genetic origin has received support (Peretz et al., 2007; Pfeifer and Hamann, 2018), the developmental trajectory of this apparently common developmental disorder is still poorly understood.

Despite the apparent specificity of the disorders characterizing each of these two conditions, developmental dyslexia and congenital amusia bear striking similarities. At the neural level, measurements of cortical thickness showed that amusic individuals have an excess of grey matter in the right inferior frontal gyrus and the right auditory cortex (Hyde et al., 2007). These observations received only partial support from voxel-based morphometry studies, which sometimes showed opposite patterns of results (Albouy et al., 2013a; Hyde, 2006; Mandell et al., 2007). Nevertheless, one possible interpretation of this increase in grey matter is that it is residual to a defect in neuronal migration, which echoes the work done in dyslexia (Galaburda et al., 1985; Galaburda and Kemper, 1979). As music and speech processing involve overlapping networks within the same cortical areas (Peretz et al., 2015), the likelihood that they would be affected together would be increased, leading to comorbidities between their respective disorders (Ramus, 2004). However, as the hypothesis of abnormal neuronal migration is very difficult to test experimentally, it must be acknowledged that a biological link between dyslexia and amusia is difficult to demonstrate empirically.

Surprisingly similar cognitive hypotheses have been independently formulated for dyslexia and amusia about a lack of conscious access to mental representations. According to these hypotheses, the phonological and musical pitch representations of these individuals would actually be intact, but less accessible. 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 and Szenkovits, 2008). This view is now being supported by a number of studies (Boets et al., 2013; Dickie et al., 2013; Ramus, 2014; Ramus and Ahissar, 2012). Similarly, the core deficit in congenital amusia has been proposed to reside in a lack of conscious access to processed pitch representations. Electrophysiological measurements revealed that the amusic brain can track and record subtle pitch variations as normal individuals do, while the outcome of these computations does not give rise to any conscious report (Moreau et al., 2013; Peretz et al., 2009). In a related vein, studies using implicit investigation approaches have reported some spared tonal structure in amusia (Albouy et al., 2013b; Omigie et al., 2012; Tillmann et al., 2012, 2016a). Both for dyslexia and amusia, these awareness impairments have been related to altered connectivity between a relatively intact auditory perceptual system and inferior frontal regions (Albouy et al., 2013a, 2015, 2019; Boets et al., 2013; Hyde et al., 2011; Kovelman et al., 2012; Ramus, 2014).

At the behavioural level, several studies suggest a partial overlap between musical deficits and phonological or speech impairments. Indeed, some musical impairments have been reported in dyslexic individuals. Conversely, fine speech processing difficulties have been reported in amusia: although amusic individuals have a normal understanding of speech and prosody in everyday life (Ayotte et al., 2002), their pitch-processing deficit might extend to subtle deficits in processing speech intonation (Hutchins et al., 2010; Liu et al., 2010; Patel et al., 2008) and emotional prosody (Lolli et al., 2015; Pralus et al., 2019) as well as processing pitch contrasts in tone language words (Liu et al., 2016; Nan et al., 2010; Tang et al., 2018; Tillmann et al., 2011; Zhang et al., 2017). Besides, a few studies reported phonological awareness impairments in a subset of amusic individuals (Jones et al., 2009; Sun et al., 2017). More generally, several empirical studies show correlations between pitch perception skills and reading level, both in typically developing children (Anvari et al., 2002; Loui et al., 2011) and in children with reading difficulties (Cogo-Moreira et al., 2013). A limitation of these findings, however, is that the effects of potentially cofounding factors such as general intelligence, music education or socio-economical level are often not taken into account (Banai and Ahissar, 2013; Carver, 1990; Fluss et al., 2009; Schellenberg, 2006).

In the light of these elements, the question arises of the co-occurrence of dyslexia and amusia, and their possible etiological link. To our knowledge, the exact relationship between the two disorders has never been directly investigated. A first exploratory study was conducted in a population of dyslexic and amusic adults. By examining their performance in literacy, phonology, and musical perception using diagnostic batteries, Couvignou et al. (2019) observed a 30% amusia rate in dyslexia and a 25% dyslexia rate in amusia. However, this first study had several limitations: in particular, the relatively small sample size (36 participants) and the fact that an adult population was considered, which induces greater environmental variability and leaves room for more robust compensatory strategies. Because they were implemented for a longer period of time, these strategies may be more prominent in adulthood, increasing the risk of masking subtle deficits. In the present study, we examined the relationships between amusia and dyslexia in a larger sample, this time focusing on children rather than adults, allowing for comparative study at an earlier stage of development.

2. Methods

2.1. Participants

Ninety-three children participated in this study, but 13 (6 dyslexic and 7 control candidates) were excluded on the basis of the inclusion phase tests described below. Four other control children with a particularly high level of musical education (more than 3 years) were further excluded to ensure group matching on this variable. The final sample included 38 children with dyslexia and 38 control children aged between 7 and 12 years. All were French native speakers who had normal hearing and normal or corrected-to-normal vision, as well as non-verbal IQ above percentile 10. They attended school regularly and none of them had any history of neurological illness or brain damage. The dyslexic children were recruited in Belgium and in France in speech therapy offices and special education institutions. The typically developing children were recruited from schools in the same cities as the dyslexic group or through personal networks. The two groups were matched on chronological age, gender, grade, musical education and parents' socio-economic status (Table 1).

Inclusion criteria required children with dyslexia to score at least 1.5 standard deviation (SD) below the grade-appropriate mean in either text or isolated word reading (accuracy or speed; Jacquier-Roux et al., 2005) and control participants to score no more than 1 standard deviation below the mean in both tests. Within the dyslexic group, eight children had comorbidities with other learning disorders: two with comorbid dyslexia and dyscalculia, five with comorbid dyslexia and attention deficit disorder (ADD), one with comorbid dyslexia, dyscalculia and ADD. In spite of this, no child scored below −2 SD on attention and oral language composite z-scores (see inclusion phase described below).

The study was conducted with approval of the local Ethics committee of the Université Libre de Bruxelles (agreement number: 034/2017). Written informed consent was obtained from the parents of each child, as well as oral agreement from each participant. All children received a "diploma" as a reward for their time and participation.

2.2. Materials and procedure

Children were tested individually in a quiet room of their school or at home, over three sessions of about 1 h each, with a few days between sessions (Table 2). The inclusion tests were administered in the first session, and the experimental tests were distributed over the other two sessions; one was devoted to the phonological tasks and the other to the musical tasks, the order of these two experimental sessions being counterbalanced between participants. Tasks were presented in a fixed order within sessions, which followed the order of the description below. Children were encouraged to take a break whenever they felt tired to maintain sustained attention throughout the tests. When conducted at home, the tests took place without the presence of parents or siblings and with minimal risk of outside distraction (i.e., sitting at a table or a desk in an isolated room). Verbal memory, phonological awareness and musical tasks were programmed under PsychoPy2 (Peirce et al., 2019). Stimuli were presented through Sennheiser HD206 headphones.

2.2.1. Inclusion phase

The *inclusion phase* consisted of several tasks that aimed at assessing attention, nonverbal intelligence, oral language, and literacy skills.

Attention skills were assessed through several tasks selected from the computerized Test for Attentional Performance (TAP, Zimmermann and Fimm 2012). In the *tonic and phasic alertness* test, a cross is displayed at irregular intervals at the midpoint of the screen, and the child has to respond to it by pushing a key-response as quickly as possible. In the tonic alertness condition (Part A), the cross appears on the screen without warning; in the phasic alertness condition (Part B), a warning tone precedes its appearance. The test consists of four blocks of 20 trials each, following an ABBA sequence. Reaction times (RTs) were recorded for each condition. A phasic attention index was calculated as follows:

Table 1
Mean age, gender (F: female), grade, music education and parents' socio-economic status (SES) as a function of group (standard deviation -SD- in parentheses). Statistics and effect sizes are shown for group differences.

	Dyslexics n = 38	Controls n = 38	T-test		Effect size Cohen's d
			t	p	
Age (years)	10.16 (1.57)	9.77 (1.43)	−1.15	.254	-.263
Gender	22 F	22 F			
Grade	4.34 (1.44)	4.13 (1.38)	- .65	.517	-.149
Music education (months)	4.30 (10.26)	6.16 (9.33)	.82	.416	.190
Parents' SES[a]	66.00 (15.93)	68.37 (15.73)	.62	.540	.150

[a] Parents' SES was estimated by averaging their Index for Individual Socio-economic Level (Genoud, 2011), calculated as follows: age – 6 x level of school completed – 4 x employment category + 55.

Table 2
Summary of the tasks administrated during the inclusion and experimental phases.

Task	Battery	Measures	Duration
INCLUSION PHASE			
Attention			20 min
Phasic alert	*Test for Attentional Performance* (TAP, Zimmermann and Fimm 2012)	• phasic attention index	
Divided attention	TAP	• visual index • auditory index	
Nonverbal intelligence			10 min
Matrices	*Raven's Colored Progressive Matrices* (Raven, 1965)	• number of correct answers	
Oral language			15 min
Receptive lexicon	French version of the *Peabody Picture Vocabulary Test* (form A; Dunn et al., 1993)	• number of correct answers	
Semantic fluency	Handmade	• number of correct answers	
Oral comprehension	*Batterie Analytique du Langage Écrit* (BALE, Jacquier-Roux et al., 2010)	• number of correct answers	
Literacy			15 min
Text reading	BALE	• words correct per minute	
Word/ pseudoword reading	ODEDYS	• accuracy • time	
Word/ pseudoword fluency	Handmade	• words/ pseudowords correct per minute	
Word/ pseudoword spelling	ODEDYS	• accuracy • time	
EXPERIMENTAL PHASE			
Phonology			50 min
Verbal memory			
Forward digit span	ODEDYS	• highest number of correctly repeated digits	
Backward digit span	ODEDYS	• highest number of correctly repeated digits	
Pseudoword repetition	*Batterie d'Évaluation du Langage Écrit* (BELEC, Mousty et al., 1994)	• number of correct answers	
Phonological awareness			
Initial syllable/ phoneme deletion	BELEC	• number of correct answers	
Syllable/ phoneme inversion	BELEC	• number of correct answers	
Acronyms	BELEC	• number of correct answers	
RAN	Handmade	• total naming time	
Music			60 min
Music exposure	*Exposure to Music in Childhood Inventory* (Cogo-Moreira and Lamont, 2018)	• score	
Music perception and memory	*Montreal Battery of Evaluation of Musical Abilities* (MBEMA, Peretz et al., 2013)	• number of correct answers for each subtest	
Pitch/time change detection	Inspired from Hyde and Peretz (2004)	• *d'* sensitivity index • decision criterion *c*	
Singing	Inspired from Dalla Bella et al. (2009) and Larrouy-Maestri et al. (2013)	• pitch deviance • time deviance • average tempo • number of omissions	

(median RT_A – median RT_B)/median RT_{total}. In the *visual selective attention* test, the child had to press the key-response as quickly as possible whenever four "X"s form a square within a 4 x 4 matrix. A total of 100 visual stimuli were presented (1 every 2 s), including 17 visual targets. In the *auditory selective attention* test, the child has to press the key as quickly as possible whenever any irregularity appears in a sequence of high (2000 Hz) and low (1000 Hz) beeps. A total of 200 auditory stimuli (1 per second) were presented, including 16 auditory targets. Both accuracy and RTs were recorded. In the *divided attention* test, the visual and the auditory tasks described above were performed simultaneously. A total of 100 visual and 200 acoustic stimuli were presented, including 17 visual and 16 acoustic targets. Both accuracy and RTs were recorded. A divided attention index was calculated as follows, separately for the visual and auditory conditions: *(number of correct responses - number of omissions - number of aberrant responses)/median RT*.

The three attention indexes were transformed into normalized z-scores based on the mean and SD of the control group, and then averaged into a composite attention z-score to ensure that all children performed above −2 SD from the mean.

Nonverbal intelligence was assessed using the Raven's Colored Progressive Matrices (Raven, 1965). In this test, children are presented with an incomplete design and six alternatives among which they must choose the one that best completes the design. The test consists of 36 visual patterns of increasing difficulty. French norms (Raven, 1998) were used to ensure that all children performed above percentile 10.

Oral language skills were assessed through three oral language tests. *Receptive lexicon* was assessed using the French version of the Peabody Picture Vocabulary Test (form A; Dunn et al., 1993). In this test, the child must choose among the four images presented to him/her the one that corresponds to the word stated by the experimenter. The test consists of 170 sheets of increasing difficulty. The stopping criterion is set at six errors out of eight consecutive trials within the application area. The score was the number of correct answers. *Semantic fluency was estimated* for two semantic categories ("sport" and "holidays"): the child had to give orally in 1 min as many words as possible pertaining to each category. The score was the total number of correct answers for both categories. *Oral comprehension* was assessed using the abbreviated version of the "Épreuve de Compréhension Syntaxico-Sémantique" (E.CO.S.SE, Lecocq, 1998) from the "Batterie Analytique du Langage Écrit" (BALE, Jacquier-Roux et al., 2010). On each of the 20 trials, the child must choose among the four presented images the one that corresponds to the sentence stated by the experimenter. Among the four images, one illustrates the situation evoked by the statement, the others represent lexical or grammatical foils. The score was the number of correct answers.

The three oral language scores were transformed into normalized z-scores based on the mean and the SD of the control group, and then averaged into a composite oral language z-score to ensure that all children performed above −2 SD from the mean.

Literacy skills were assessed through four types of tasks. Text reading was assessed using the BALE (Jacquier-Roux et al., 2010). Word and pseudoword reading as well as word and pseudoword spelling were assessed by the French battery "Outil de Dépistage des Dyslexies" (ODEDYS, Jacquier-Roux et al., 2005). In the *text reading* test, children were asked to read aloud the text "Monsieur Petit" for 1 min. The number of words read without error was scored. To evaluate *word/pseudoword reading*, children were administered three lists of 20 items each: irregular words, regular words and pseudowords. They were asked to read them aloud as quickly and as accurately as possible. Both accuracy and speed were scored. In the *word and pseudoword reading fluency* task, children were presented with a list of 160 isolated items. They were asked to read aloud as many of them as possible without error, in 1 min. The first list was composed of seven-letters words from diverse grammatical categories (verb, noun, adjective, adverb), presented in a fixed random order; 100 of them were frequent (i.e. standard frequency index > Q3, Manulex database) (Lété et al., 2004), 60 were rare (standard frequency index < Q1); 120 were regular, 40 were irregular. The second list was composed of pseudo-words created from the words by trigram recombination with Lexique Toolbox (New et al., 2001). The score was the number of words/pseudowords correct per minute. To examine *word and pseudoword spelling*, three lists of 10 items each were dictated to the children: irregular words, regular words and pseudowords. Both accuracy and speed were scored.

2.2.2. Experimental phase

The *experimental phase* aimed at assessing both verbal and musical skills.

As regards verbal skills, we examined verbal memory, phonological awareness and rapid access to phonological information.

Short-term and working memory was assessed through two digit span tests taken from the ODEDYS battery. Children were asked to repeat sequence of digits of increasing length (2–9 items), in forward and backward order. The stopping criterion was set at two consecutive failures in a series of sequences of the same length. We scored forward and backward span, i.e., the highest number of correctly repeated digits for each condition. *Phonological memory* was assessed through the nonword repetition tests of the "Batterie d'Évaluation du Langage Écrit" (BELEC, Mousty et al., 1994) . Children were instructed to repeat nonwords of increasing length (1–5 syllables) as accurately as possible. Five series of four items each were presented under two conditions: the pseudowords' syllables had either a simple structure (consonant-vowel, CV condition) or a complex one (consonant-consonant-vowel, CCV condition). The number of words repeated without error was scored.

Three *phonological awareness* tasks were selected from the BELEC (Mousty et al., 1994). In the initial syllable/phoneme deletion tests, children were required to repeat CVCV pseudowords without the initial syllable (16 items) as well as to repeat CVC (16 items) and CCV (10 items) monosyllables without the initial phoneme. Four practice trials were given before each condition. The score was the total number of correct answers. In the syllable/phoneme inversion tests, children were asked to swap the syllables of CVCV pseudowords (10 items) as well as the phonemes of VC or CV monosyllables (10 items) and to produce the resulting pseudoword. Four practice trials were given before each condition. The score was the total number of correct answers. In the tests of *acronyms*, children were verbally presented with 16 pairs of words and had to produce a new word resulting from the merging of the initial phonemes of the two words. Four practice trials were given before each condition. The score was the number of correct answers.

Rapid access to phonological information was evaluated through a Rapid Automatized Naming (RAN) task, in which children named series of 50 items (either objects or colors) as fast as possible. Each RAN test was administered twice with different sheets. The score was the sum of total naming time for both sheets of each test.

As regards musical skills, we evaluated music exposure and assessed music perception and memory as well as singing.

Children completed orally a French version of the *Exposure to Music in Childhood Inventory* (Cogo-Moreira and Lamont, 2018). This 14-item questionnaire is designed to capture their amount and type of exposure to music activities, including exposure to multimedia, the internet and television alongside more conventional elements of family background and activities at school. Each item of the questionnaire is responded to with a scale ranging from 0 to 4, resulting in a score range of 0–30 such that higher scores represent higher music exposure.

Music perception and memory was assessed using the *Montreal Battery of Evaluation of Musical Abilities* (MBEMA, Peretz et al., 2013), which involves five tests measuring scale, contour, interval, rhythm and memory of unfamiliar but conventional melodies. Each test comprises two practice trials with feedback followed by 20 experimental trials and uses the same pool of 20 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 *Memory* task requires children to recognize a melody as having been presented earlier during the session or not. Amusia was diagnosed when an individual performed two SD below the mean of controls in the global score (mean score of the 5 tests) or in the melodic composite score (mean score of the first 3 tests: Scale, Contour and Interval).

In the *pitch/time change detection tests*, children were presented with monotonic and isochronous sequences of five tones (i.e., constant pitch and intertone interval) and were required to detect when the fourth tone was displaced either in pitch or time (Hyde and Peretz, 2004). In the standard sequence, all tones were 100 ms long, played at the pitch level of C6 (1047 Hz) with an intertone interval of 350 ms. In the pitch-altered sequences, the fourth tone was displaced by one of five pitch distances upward or downward from C6, ranging from 25 to 300 cents. In the time-altered sequences, the fourth tone was displaced by one of five temporal increments earlier or later than its isochronous position, ranging from 8 to 16% of the intertone interval. Pitch and time change detection were tested in two separate blocks. Children received 10 practice trials with feedback after each trial, which they could replay in case of failure. Experimental trials were composed of 60 randomised sequences (30 standard sequences, 3 of each 10 altered sequences).

Singing was evaluated off-line: children were recorded singing the French version of the popular tune "Happy Birthday", once with usual lyrics and once on the syllable/la/. No particular starting note was given to let the participant choose his/her comfortable range. Children were encouraged to sing at an easy pace as singing too fast has negative effect on pitch accuracy (Dalla Bella et al., 2007). Of the records, those of three dyslexic children were unusable, incomplete or missing: the records of one dyslexic child were too damaged to be used, one dyslexic child with comorbid ADD did not manage to sing the tune without the lyrics and one dyslexic child refused to sing despite our encouragement. The analyses were thus carried out on 147 records representing data from 38 control children and 36 dyslexic children, one of whom had incomplete data. As done in previous studies (Larrouy-Maestri et al., 2013; Larrouy-Maestri and Morsomme, 2014), we took into account 21 out of 25 notes of the tune, ignoring the repeated ones (Fig. 1). Each note was associated with one syllable. Acoustical analyses were performed on the vowels, which are the best targets given that they carry the maximum of voicing and mark the onset of musical tones (Murayama et al., 2004; Sundberg and Bauer-Huppmann, 2007). In a first step, the auditory signal was segmented using Praat Software (Boersma and Paul, 2001). Markers were manually placed on the spectrogram, where the Fundamental frequency (F_0) was the most stable (i.e., avoiding the attacks and the glides between notes) and at the onset of each vowel. The mean of the F_0 within the stable part of the vowel was used to measure pitch height. The onset of vowels was considered as the note onset time. For each production, *pitch* and *time deviance* were estimated by measuring the absolute difference between the size of intervals performed (in cents or in ms) and the standard size based on the musical score (Fig. 1). The *average tempo* of the singing (in bpm) was calculated on the basis of the performance length. We also measured the *number of omissions*, which were quite frequent.

3. Results

3.1. Group comparisons

In the inclusion tasks, no significant group difference was observed for attention skills or oral language tests. Yet dyslexic children scored significantly lower than controls in all reading tasks and on most measures of phonology and orthography (see Table 3). They also differed slightly in nonverbal intelligence. Therefore, when appropriate, the contribution of this variable was partialed out in the relevant analyses.

Music exposure. Despite the fact that the two groups were matched in terms of music education (Table 1), dyslexic children reported a slightly lower level of exposure to music than controls (Table 3). Again, when appropriate, the contribution of this variable was partialed out in the relevant analyses.

Music perception and memory. Dyslexic children scored significantly lower than controls in all subtests of the MBEMA (Table 3). At an individual level, 13 of them performed under the criterion for amusia diagnosis (global or composite melodic score under −2 SD from the mean of controls) against two controls, eight showing a concomitant deficit in the rhythm test. Such a proportion of 13 out of 38 (34%) is a significantly higher rate than the one observed in the control group (5%, Yates's $\chi^2 = 8.31$, $p = .004$) and is also significantly higher than what would be expected from the population prevalence of amusia (between 1.5 and 4%: 0.57 out of 38 expected according to the minimal value of 1.5%, Yates' $\chi^2 = 11.72$, $p < .001$; 1.52 out of 38 expected according to the maximal value of 4%, Yates' $\chi^2 = 9.35$, $p = .002$). As shown in Fig. 2, five of the 13 dyslexic children with amusia were already diagnosed with other comorbid disorders: one with comorbid dyslexia and dyscalculia, three with comorbid dyslexia and ADD, and one with comorbid dyslexia, dyscalculia and ADD. Visual inspection of the variability of the RTs during the task suggested that the poor performance of the children with ADD was not due to a lack of vigilance (see supplementary material: Fig. s1).

Pitch/time change detection. Pitch and time change detection performance was assessed using Signal Detection Theory (Macmillan and Creelman, 2004). We calculated *d'* sensitivity index and decision criterion *c* as $d' = z(H) - z(FA)$ and $c = -0.5 * [z(H) + z(FA)]$ for each participant as a function of the type and level of change. Hits and false alarms were defined as follows: $H = p$ (response = change | stimulus = change) and $FA = p$ (response = change | stimulus = no change).

A 2 (group) x 2 (type of change: pitch; time) x 5 (level of change: from 25 to 300 cents for pitch, from 8 to 16% of intertone interval for time) mixed-design ANOVA was computed on these two measures. Regarding the ANOVA ran on *d'*, it revealed significant main effects of group, $F(1, 74) = 5.44$, $p = .022$, $\eta^2_p = .068$, type of change, $F(1, 74) = 125.68$, $p < .001$, $\eta^2_p = .629$, and level of change, $F(4, 296) = 50.83$, $p < .001$, $\eta^2_p = .407$ but no significant interaction (all $Fs < 1$ except for group x level of change: $F(4, 296) = 2.37$, $p = .052$). As shown in Fig. 3, the dyslexic group scored significantly lower than the control group for both types of change and for most levels, although the time change detection task turned out to be more difficult than the pitch one, and performance increased with the level of change. Regarding the ANOVA ran on decision criterion *c*, we observed significant main effects of type of change, $F(1, 74) = 20.94$, $p < .001$, $\eta^2_p = .221$ and level of change, $F(4, 296) = 50.74$, $p < .001$, $\eta^2_p = .407$ but no significant effect of group or interaction (all $Fs < 1$ except for group x type of change: $F(1, 74) = 2.44$, $p = .123$ and group x level of change: $F(4, 296) = 2.36$, $p = .053$). In line with the *d'* analyses, decision criteria were higher in the time change detection task than in the pitch one and decreased with the level of change.

As a group, the 13 children with comorbid dyslexia and amusia

Fig. 1. Score of the tune "Happy Birthday" with the number of notes used for calculating pitch and time deviance. Adapted, with permission, from Larrouy-Maestri et al. (2013).

Table 3
Mean scores for inclusion and experimental measures depending on the group (Standard deviation -SD- in parentheses). Statistics and effect sizes are shown for group differences.

	Dyslexics n = 38	Controls n = 38	T-test		Effect size Cohen's d
			t	*p*	
INCLUSION PHASE					
Attention					
Phasic alert (index)	.04 (.11)	.06 (.10)	.70	.486	.161
Divided attention (visual index)	.01 (.00)	.01 (.01)	1.40	.166	.321
Divided attention (auditory index)	.02 (.01)	.02 (.01)	1.06	.291	.244
Nonverbal intelligence					
Matrices (/36)	29.26 (3.62)	30.97 (2.70)	2.34	.022	.536
Oral language					
Receptive lexicon (score)	109.58 (14.36)	114.66 (14.94)	1.51	.135	.347
Semantic fluency (score)	22.26 (8.75)	24.90 (7.10)	1.44	.154	.330
Oral comprehension (/20)	17.32 (1.69)	17.61 (1.64)	0.76	.451	.174
Literacy					
Text reading (wpm)	63.29 (37.72)	133.03 (36.97)	8.14	<.001	1.868
Word/pseudoword reading					
IW (/20)	7.08 (4.50)	14.95 (3.88)	8.16	<.001	1.873
IW (s)	65.71 (43.56)	23.18 (8.72)	−5.90	<.001	−1.354
RW (/20)	13.42 (4.31)	18.97 (1.00)	7.74	<.001	1.775
RW (s)	59.03 (42.39)	22.26 (8.28)	−5.25	<.001	−1.204
PW (/20)	10.74 (3.10)	16.47 (2.05)	9.52	<.001	2.183
PW (s)	59.32 (39.34)	29.00 (9.91)	−4.61	<.001	−1.057
Word/pseudoword fluency					
W (wpm)	17.05 (11.53)	50.74 (21.35)	8.56	<.001	1.963
PW (pwpm)	11.79 (7.20)	32.66 (13.38)	8.47	<.001	1.942
Word/pseudoword spelling					
IW (/10)	2.63 (2.21)	6.47 (2.41)	7.24	<.001	1.660
IW (s)	93.74 (35.65)	83.40 (33.37)	−1.31	.196	-.300
RW (/10)	5.40 (3.05)	8.84 (1.59)	6.19	<.001	1.420
RW (s)	85.71 (36.17)	68.42 (30.23)	−2.26	.027	-.519
PW (/10)	6.47 (2.79)	8.92 (1.15)	5.01	<.001	1.148
PW (s)	98.76 (38.66)	88.92 (39.20)	−1.10	.274	-.253
EXPERIMENTAL PHASE					
Phonology					
Verbal memory					
Forward digit span (score)	4.79 (1.17)	5.26 (.89)	1.99	.050	.456
Backward digit span (score)	3.45 (.80)	3.74 (1.03)	1.37	.175	.314
PW repetition (/40)	24.66 (3.25)	27.47 (3.62)	3.57	<.001	.818
Phonological awareness					
Initial S/P deletion (/42)	36.74 (6.58)	40.66 (1.91)	3.53	<.001	.810
S/P inversion (/20)	16.63 (3.87)	18.92 (1.44)	3.42	.001	.785
Acronyms (/16)	12.51 (4.08)	14.63 (1.34)	3.03	.003	.709
RAN (s)	236.45 (67.88)	184.08 (41.93)	−4.05	<.001	-.928
Music					
			2.25	.027	.517

Table 3 (*continued*)

	Dyslexics n = 38	Controls n = 38	T-test		Effect size Cohen's d
			t	*p*	
Music exposure (/30)	11.29 (3.95)	13.18 (3.36)			
MBEMA					
Scale (/20)	14.68 (2.87)	16.45 (1.75)	3.24	.002	.742
Contour (/20)	15.34 (2.97)	17.37 (1.67)	3.67	<.001	.841
Interval (/20)	14.92 (3.19)	16.76 (2.15)	2.95	.004	.677
Rhythm (/20)	15.26 (3.61)	17.45 (2.45)	3.09	.003	.709
Memory (/20)	15.74 (3.11)	17.61 (2.06)	3.09	.003	.709

Note. Wpm = words correct per minute, PWpm = pseudowords correct per minute; IW = Irregular Words, RW = Regular Words, PW = Pseudo Words, W = Words, S = Syllable, P = Phoneme.

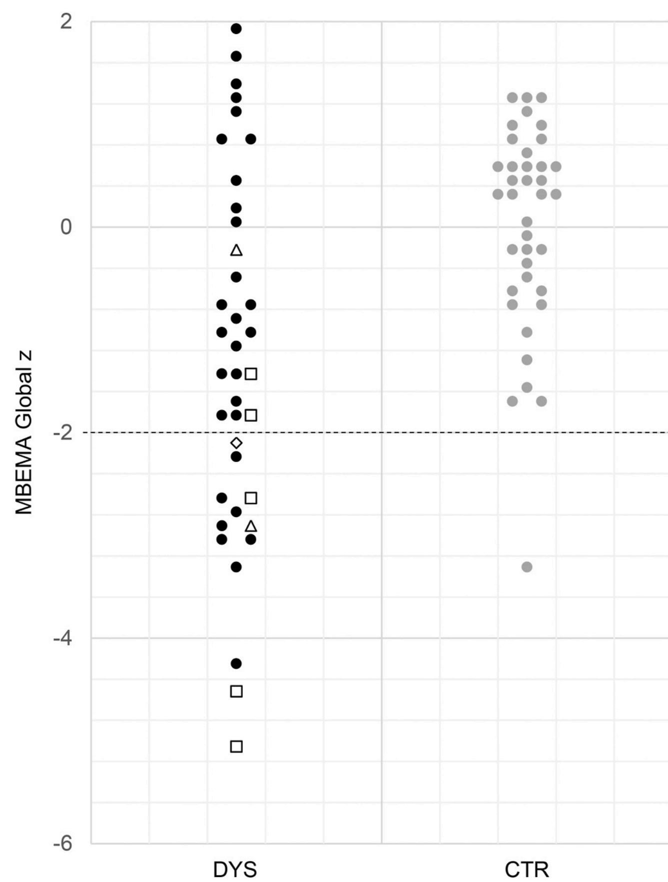

Fig. 2. Distribution of dyslexic and control participants along their global composite z-scores (computed from the mean and standard deviation of the controls) in the MBEMA. The two children with comorbid dyslexia and dyscalculia are represented with triangles, the five with comorbid dyslexia and ADD with squares and the one with comorbid dyslexia, dyscalculia and ADD with a diamond. The dashed line represents the diagnostic threshold for congenital amusia. Considering the melodic composite score, one additional control child performed under the threshold.

showed poor performance in both the pitch (mean *d*': 1.66 ± 1.48 SD vs. 3.20 ± 1.54 SD for controls and 3.15 ± 2.26 SD for dyslexic children without amusia; mean c: 0.31 ± 0.61 SD vs. 0.03 ±0 .70 SD for controls and −0.02 ±0 .50 SD for dyslexic children without amusia) and time conditions (mean *d*': 0.64 ± 0.45 SD vs. 1.45 ± 1.01 for controls and

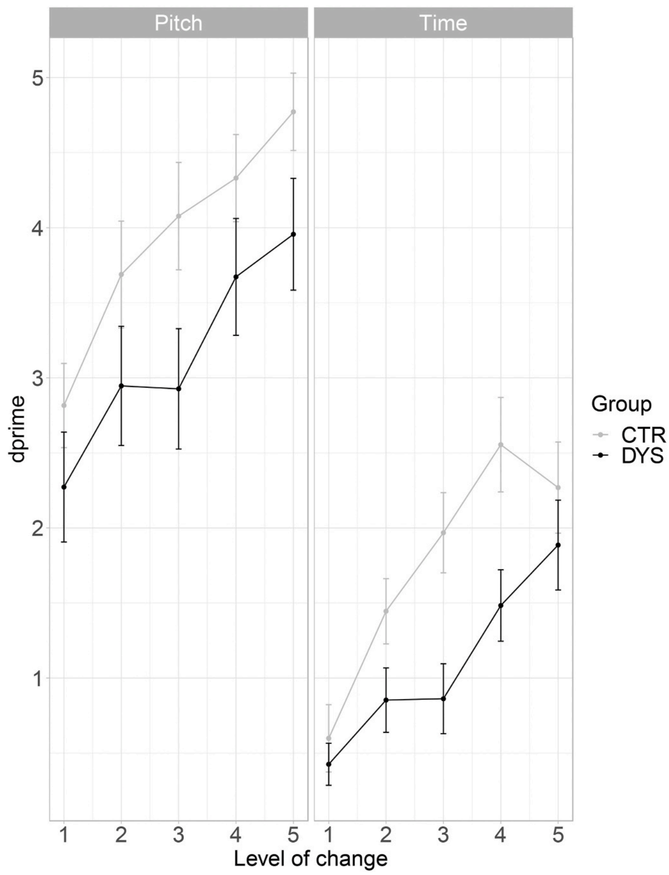

Fig. 3. Performance of the dyslexic and control groups in the pitch and time discrimination tasks, as a function of change in pitch and intertone interval, respectively. Error bars represent standard errors.

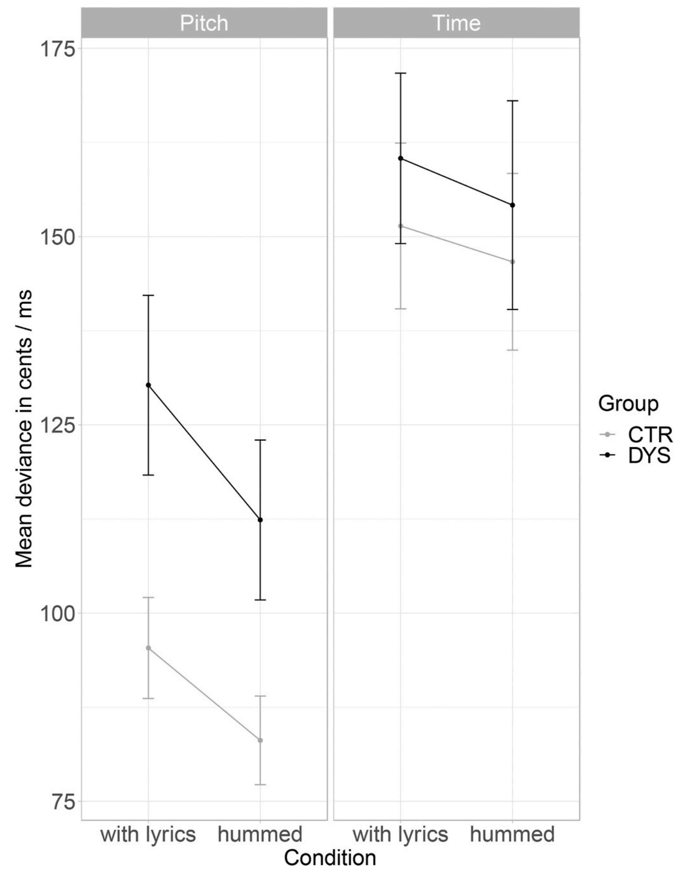

Fig. 4. Mean pitch and time deviance of the dyslexic and control groups during the singing task, as a function of condition. Error bars represent standard errors.

1.01 ± 0.81 SD for dyslexic children without amusia; mean c: 0.20 ± 0.29 SD vs. 0.36 ± 0.56 for controls and 0.21 ± 0.42 SD for dyslexic children without amusia). Nevertheless, one of them was unimpaired on the pitch domain (i.e., scored above −1 SD from the mean of controls), three on the time domain and three on both, suggesting that their low performance on the MBEMA cannot be explained solely by poor discrimination skills. A 2 (group: children with comorbid dyslexia and amusia; dyslexic children without amusia) x 2 (type of change: pitch; time) mixed design ANOVA ran on *d'* showed significant main effects of group, $F(1, 36) = 4.71$, $p = .037$, $\eta^2_p = .116$ and type of change, $F(1, 36) = 28.98$, $p < .001$, $\eta^2_p = .446$, but no significant interaction, $F(1, 36) = 3.70$, $p = .062$.

Singing. Separate 2 (group) x 2 (condition) mixed-design ANOVAs were computed on each of the four measures collected: pitch deviance, time deviance, average tempo, and number of omissions. Regarding pitch deviance, the ANOVA revealed significant main effects of group, $F(1, 71) = 6.38$, $p = .014$, $\eta^2_p = .082$, and condition, $F(1, 71) = 11.76$, $p < .001$, $\eta^2_p = .142$, but no significant interaction ($F < 1$). As shown in Fig. 4, the dyslexic group presented a significantly higher mean pitch deviance than the control group. Besides, singing in tune turned out to be more difficult with than without lyrics. In contrast, time deviance analysis revealed no main significant effect of group, $F < 1$ or condition, $F(1, 71) = 1.20$, $p = .277$, and no significant interaction, $F < 1$ (Fig. 4). The average tempo was similar between groups and conditions, without interaction (all *F*s < 1). Finally, regarding the number of omissions, there was a main effect of group, $F(1, 71) = 6.3$, $p = .014$, $\eta^2_p = .082$, but no main effect of condition or interaction (both *F*s < 1). Dyslexic children committed more omissions than controls (mean: 2.44 ± 2.78 SD vs. 1.03 ± 1.63 SD, respectively) regardless of the condition.

As expected, most of the children with comorbid dyslexia and amusia exhibited poor singing, especially on the pitch dimension (mean pitch deviance: 139.23 ± 53.63 SD vs. 89.23 ± 35.96 for controls and 114.56 ± 73.22 for dyslexic children without amusia, mean time deviance: 188.10 ± 86.87 SD vs. 149.03 ± 67.61 for controls and 138.80 ± 50.27 for dyslexic children without amusia, mean tempo: 99.68 ± 13.56 SD vs. 108.18 ± 16.28 for controls and 113.79 ± 18.95 for dyslexic children without amusia, mean number of omissions: 3.5 ± 3.25 SD vs. 1.03 ± 1.63 for controls and 1.85 ± 2.35 for dyslexic children without amusia). They were also one of the few children to commit errors on the melodic contour (i.e., the direction of the produced interval was opposite to that of the musical score; mean number: 0.96 ± 1.07 SD vs. 0.18 ± 0.41 SD for controls and 0.54 ± 1 for dyslexic children without amusia). As mentioned in the method section, one of these children (with comorbid dyslexia, amusia and TDA) was unable to sing the tune without the lyrics. Interestingly, however, two of these children were unimpaired on the pitch domain (i.e., scored above −1 SD from the mean of controls), four on the time domain and four on both, which suggests a partial dissociation between perception and production.

3.2. Relationships between literacy, phonological and musical components

The results of all tasks were transformed into standardized z-scores based on the means and standard deviation of the control group. Six theory-driven components were then computed: a *literacy accuracy component* (average of the accuracy scores on text reading, irregular word reading, regular word reading, pseudoword reading, word fluency, pseudoword fluency tasks, irregular word spelling, regular word spelling and pseudoword spelling tasks), a *literacy speed component* (average of the speed scores on irregular word reading, regular word reading, pseudoword reading, irregular word spelling, regular word spelling and pseudoword spelling tasks), a *phonological awareness/RAN component* (average of scores on initial syllable/phoneme deletion, syllable/phoneme inversion, acronyms and RAN tasks), a *verbal memory component* (average of scores on forward digit span, backward digit span and pseudoword repetition tasks), a *pitch processing component* (average of the scores on MBEMA Scale, MBEMA Contour, MBEMA Interval, pitch change detection, pitch singing deviance as well as the MBEMA memory score with a 0.5 loading), and a *time processing component* (average of MBEMA Rhythm, time change detection, time singing deviance and the MBEMA memory score with a 0.5 loading). Because the MBEMA incidental memory test involved both pitch and time processing, we allowed this variable to cross-load equally on pitch and time dimensions.

As a first step, we computed correlations between literacy, phonological and musical skills, as represented by our composite variables (Table 4). Since a large number of correlations were calculated, we corrected the analyses for multiple comparisons, applying a Bonferroni correction on these 15 measures. As expected from a large body of research on literacy, literacy accuracy, literacy speed and phonological awareness/RAN components were highly correlated to each other across groups, but also, for the most part, within each group.

The two musical components were also highly associated. Besides, as illustrated in Fig. 5, a number of inter-domain correlations were significant across groups, in particular, literacy accuracy and phonological awareness/RAN with pitch processing. As illustrated in Table 4, these associations were independent from potential cofounding factors such as chronological age, parents' SES, music education, music exposure, nonverbal intelligence or attention skills. However, none of them reached significance when carried out by group. In fact, these inter-domain correlations only reflected between-group differences or were carried by the few individuals with difficulties in both domains, thus suggesting that this is not a general relationship.

We then carried out backward multiple regressions analyses to investigate whether musical skills could be independent predictors of participants' literacy scores. Literacy accuracy and literacy speed were successively entered as dependent variables, with phonological awareness/RAN, verbal memory, pitch processing, time processing, chronological age, parents' SES, music education, music exposure, composite attention z-score and nonverbal intelligence as predictors. As shown in Table 5, the pitch processing component was a significant predictor of literacy accuracy across groups and of literacy speed within the dyslexic group. In contrast, the time processing component did not predict any measures of literacy.

4. Discussion

This study investigated for the first time the degree of association between developmental dyslexia and congenital amusia in children. Dyslexic children and matched controls were behaviorally assessed with the respective diagnostic batteries for both disorders, measuring reading and phonological skills on the one hand, and musical perception and production on the other.

We found a significant overlap (about 34%) between dyslexia and amusia, which is comparable to commonly reported rates of comorbidity between dyslexia and other learning disabilities such as developmental language disorder or ADD. In addition, we observed moderate to strong positive correlations between pitch processing skills, literacy accuracy and phonological skills after partialing out potential cofounding factors such as age, parents' SES, music education, general intelligence and attention skills. These correlations, however, where mostly driven by group differences. Multiple regression analyses indicated that pitch processing skills were a significant predictor of literacy accuracy across groups and of literacy speed within the dyslexic group.

We will first examine several possible explanations for an increased comorbidity between dyslexia and amusia, at both the cognitive and neural levels. We will then consider the cognitive profile of comorbid cases. Finally, after discussing some of the limitations of this study, we

Table 4
Partial linear correlations between the composite variables (a) across groups, (b) within the dyslexic group (c) within the control group. Correlations are reported after partialing out age, parents' SES, music education, music exposure, composite attention z-score and nonverbal intelligence. *P*-values are reported after being corrected for multiple comparisons (Bonferroni correction). In bold: significant correlations.

(a)

Component variable	Literacy speed		Phonological awareness/RAN		Verbal memory		Pitch processing		Time processing	
	r	*p*	r	*p*	r	*p*	r	*p*	r	*p*
Literacy accuracy	**.70**	**<.001**	**.53**	**<.001**	.29	.302	**.41**	**.014**	.31	.197
Literacy speed			**.74**	**<.001**	.26	.574	.28	.385	.17	1.000
Phonological awareness/RAN					.33	.129	**.45**	**.004**	.33	.141
Verbal memory							.28	.453	.20	1.000
Pitch processing									**.77**	**<.001**
Time processing										

(b)

Component variable	Literacy speed		Phonological awareness/RAN		Verbal memory		Pitch processing		Time processing	
	r	*p*	r	*p*	r	*p*	r	*p*	r	*p*
Literacy accuracy	.48	.140	**.61**	**.008**	.11	1.000	.17	1.000	.02	1.000
Literacy speed			**.74**	**<.001**	.15	1.000	.04	1.000	-.12	1.000
Phonological awareness/RAN					.31	1.000	.35	1.000	.11	1.000
Verbal memory							.28	1.000	.21	1.000
Pitch processing									**.79**	**<.001**
Time processing										

(c)

Component variable	Literacy speed		Phonological awareness/RAN		Verbal memory		Pitch processing		Time processing	
	r	*p*	r	*p*	r	*p*	r	*p*	r	*p*
Literacy accuracy	**.71**	**<.001**	-.10	1.000	.12	1.000	.15	1.000	.05	1.000
Literacy speed			-.05	1.000	.06	1.000	.03	1.000	-.04	1.000
Phonological awareness/RAN					.14	1.000	.05	1.000	.29	1.000
Verbal memory							-.04	1.000	-.10	1.000
Pitch processing									.518	.071
Time processing										

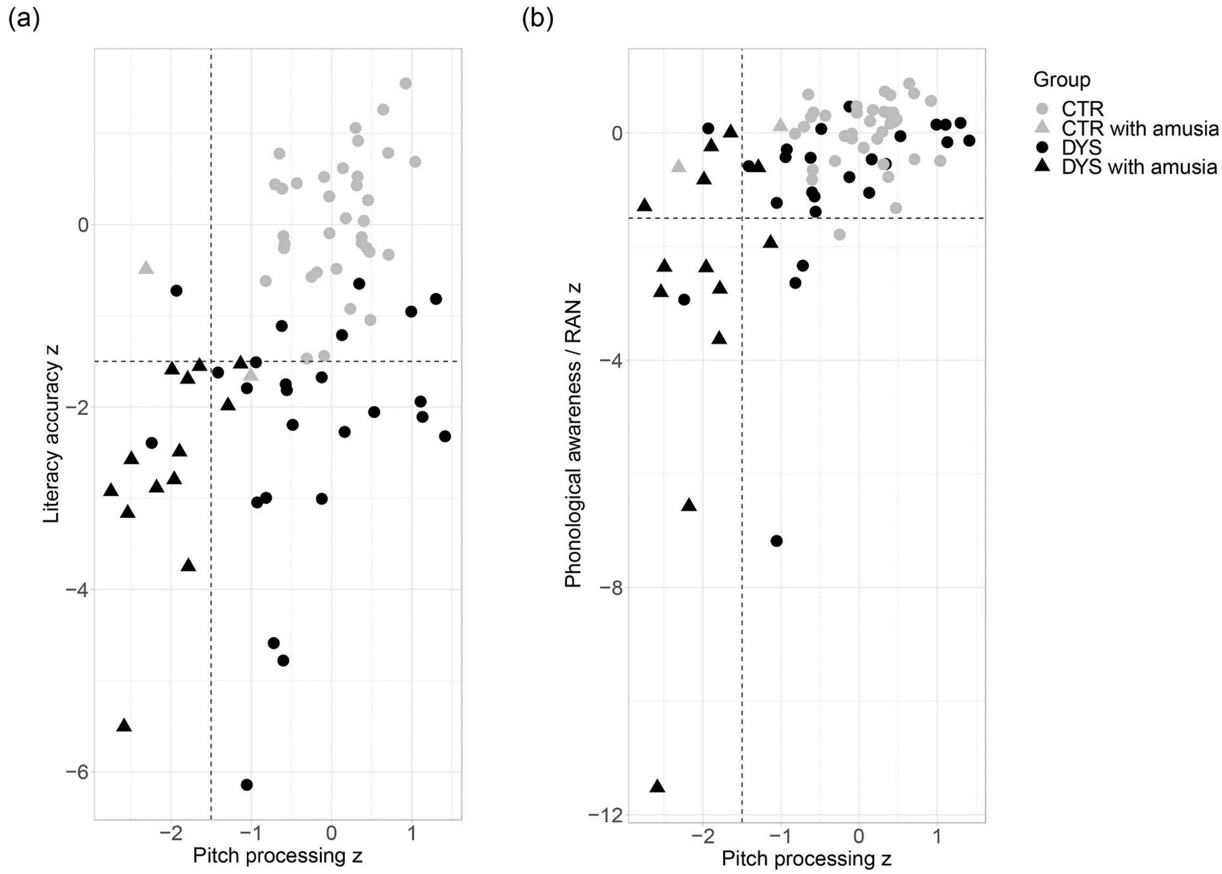

Fig. 5. Distribution of individual composite z-score along (a) *literacy accuracy* and *pitch processing* factors, (b) *phonological awareness/RAN* and *pitch processing* factors. The horizontal and vertical lines indicate the −1.5 standard deviation threshold.

Table 5
Best predictor models retained by the backward multiple regression analyses (a) across groups, (b) within the dyslexic group (c) within the control group. Stepping method criteria with *p*-value was used (entry $p = .05$; removal $p = .10$).

(a)

Dependent variable	Adjusted R^2	ANOVA		Predictors	B	Beta	*t*	*p*
		F	p					
LITERACY ACCURACY	.49	22.11	<.001	Phonological awareness/RAN	.38	.47	4.52	<.001
				Pitch processing	.35	.28	2.65	.010
				Attention	.38	.18	2.00	.049
LITERACY SPEED	.63	39.76	<.001	Phonological awareness/RAN	.69	.72	9.49	<.001
				Age	.17	.16	2.02	.047
				Attention	.43	.17	2.16	.035

(b)

Dependent variable	Adjusted R^2	ANOVA		Predictors	B	Beta	*t*	*p*
		F	p					
LITERACY ACCURACY	.69	25.88	<.001	Phonological awareness/RAN	.23	.50	4.67	<.001
				Age	.26	.43	4.24	<.001
				Nonverbal intelligence	.14	.19	1.87	.071
LITERACY SPEED	.70	27.17	<.001	Phonological awareness/RAN	.70	.79	7.02	<.001
				Pitch processing	-.39	-.26	−2.33	.027
				Age	.43	.36	3.65	<.001

(c)

Dependent variable	Adjusted R^2	ANOVA		Predictors	B	Beta	*t*	*p*
		F	p					
LITERACY ACCURACY	.53	19.22	<.001	Age	.33	.62	4.82	<.001
				Nonverbal intelligence	.17	.23	1.80	.082
LITERACY SPEED	.53	19.41	<.001	Age	.31	.58	4.19	<.001
				Attention	.31	.26	1.89	.068

will address its implication for future research and practices.

4.1. Partial overlap between developmental dyslexia and congenital amusia

A first result of the present study is that about 34% of the dyslexic children in the sample (13 out of 38) also meet diagnostic criteria for congenital amusia. This proportion is significantly higher than both the rate expected based on the prevalence of amusia in the general population (1.5–4%) and the rate observed in the matched control sample (2 out of 38, i.e., about 5%). With regard to the latter point, it can be pointed out that one of the two control children with amusia, although meeting the inclusion criteria, had the lowest literacy accuracy score among controls (below the −1.5 standard deviation threshold).

The present result is consistent with previous data collected on adults (Couvignou et al., 2019), which suggested a 30% amusia rate in dyslexia. The rate of comorbidity observed in the present study is even slightly higher, which may be explained by the larger sample size and/or by the fact that the adult population could be more inclined to use efficient compensatory strategies that may mask potential deficits. In addition, as mentioned by Couvignou et al. (2019), the adult sample considered was highly educated and socially privileged, therefore potentially more resourceful and with better access to remediation, thus decreasing the likelihood of comorbidity.

Nevertheless, it should be noted that although dyslexia and amusia seem to be frequently comorbid, their association is not systematic: pure amusia exists just like pure dyslexia. As reported in previous studies (Bishop-Liebler et al., 2014; Weiss et al., 2014), having dyslexia is not incompatible with high musical skills. Likewise, one dyslexic child in our sample demonstrated an excellent musical ear (100% correct answers in the MBEMA) even though she had no formal musical education. Although group differences were observed on all musical measures, the majority of dyslexic children performed above the pathological threshold on musical tests. Thus, it may be relevant in future studies to distinguish pure cases of either of these disorders from comorbidities.

4.2. Explanatory hypotheses

There are at least three non-exclusive explanatory hypotheses that could account for enhanced comorbidity between dyslexia and amusia (Fig. 6): a shared underlying cognitive deficit, a causal relationship between musical and phonological deficits and a common genetic risk factor inducing neural disruptions in specific areas (henceforth, H1, H2 and H3, respectively).

According to H1, amusia and dyslexia could share a common underlying deficit. Indeed, if a same cognitive function is altered and affects both the musical and linguistic domains, it would contribute to increase the co-occurrence of both disorders. Several cognitive skills are candidate: the capacity to discriminate auditory perceptive units, to integrate them into short-term memory, to pay attention to these units, or to make mental operations on these. Most of these functions have been investigated, but for each disorder separately, on various dimensions which are not directly comparable, and with highly variable paradigms, stimuli, and tasks. It would be worthwhile to examine these skills in parallel in a more systematic way for each domain of specificity of the two disorders.

The test batteries used in the present study were chosen for their diagnostic properties and do not distinguish between the cognitive skills listed above as candidates. Still, we can notice that the cases of comorbidity scored among the lowest in the *phonological awareness/RAN* component (eight cases out of 13 under the −1.5 threshold). This observation had already been made by Couvignou et al. (2019) in their adult study and is consistent with the results of previous works reporting weak phonological awareness in a significant proportion of amusic individuals (Jones et al., 2009; Sun et al., 2017). As argued in the introduction, both amusia and dyslexia can be conceptualized as disorders of conscious access to mental representations (Loui et al., 2011; Peretz et al., 2009; Ramus and Ahissar, 2012). Accordingly, cases of comorbidity might arise from an access deficit affecting the processing of both pitch and speech sounds.

According to H2, amusia and dyslexia would tend to coexist because of a relation of causality between musical and language-related skills. In the same way as musical expertise may facilitate language processing, as proposed for instance by Besson et al. (2011) and Patel (2011), the presence of congenital musical difficulties could alter language processing. Reciprocally, it could be that early difficulties in speech processing favour the development of musical disorders. Several studies show correlations between music perception abilities and reading level in children (e.g., Anvari et al., 2002; Banai and Ahissar, 2013; Cogo-Moreira et al., 2013), but they do not allow to establish a link of causality.

Our own observations suggest that some links between the two

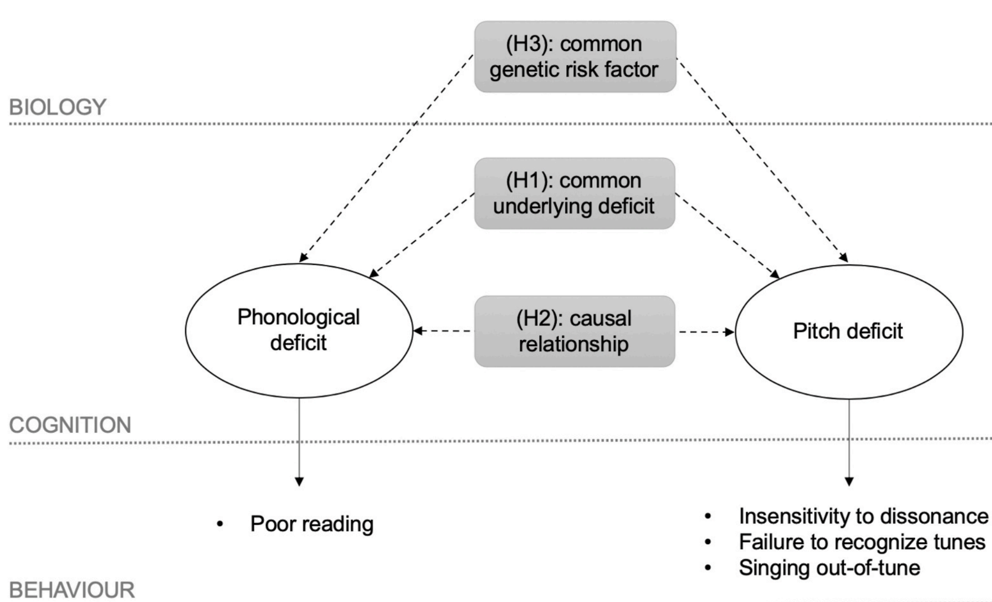

Fig. 6. Three non-exclusive explanatory hypotheses for enhanced comorbidity between dyslexia and amusia.

domains are resistant to the control of potential confounding variables. In particular, even after controlling for age, parents' SES, music education, music exposure, composite attention z-score and nonverbal intelligence, the pitch processing component was correlated with both phonological skills and literacy accuracy. Although these links seemed to reflect group differences more than a genuine relationship between the two domains, pitch processing skills still predicted literacy accuracy across groups and literacy speed within the dyslexic group.

In contrast, the time processing component showed no association with measures of literacy or phonology. This is somewhat inconsistent with a growing set of data linking musical rhythm to speech/language processing (for a recent review, see Ladányi et al., 2020). Still, time processing is less directly related to the type of congenital amusia we have examined here, namely *tone deafness*, which is mainly due to a defect in pitch perception and memory and is therefore distinct from *beat deafness* (Phillips-Silver et al., 2011, 2013). The tasks we employed were therefore mainly focused on pitch processing, and potentially less complete and less demanding (with the exception of the time change detection task) with respect to the temporal dimension. Accordingly, we did not assess beat perception or synchronization skills, which were reported to be weaker in dyslexia (e.g., Colling et al., 2017; Goswami et al., 2013; Thomson and Goswami, 2008). More generally, the outcomes of these correlation/regression analyses should be interpreted with caution, as the use of composite scores may lead to unintended confusion of the variables. In particular, the constituent sub-tests of the musical components were not always correlated with each other (see supplementary material: Table s1).

In any case, intervention studies and longitudinal designs would be more appropriate for testing causality. However, to date, few intervention studies have been well enough designed to allow drawing reliable conclusions (Cogo-Moreira et al., 2012; but see Flaugnacco et al., 2015), and very few longitudinal studies have attempted to trace the comparative developmental curve of music and language skills. If some transfer effects existed between the two domains, they are likely to be small (Gordon et al., 2015; Sala and Gobet, 2017). Therefore, this hypothesis still lacks supporting evidence.

Alternatively, according to H3, associations may arise at the neural level without being underpinned by a cognitive link between the musical and the phonological deficit. Indeed, a common genetic risk factor could induce neural disturbances in specific areas, leading to the simultaneous disruption of the development of several yet independent cognitive functions (Galaburda et al., 2006; Ramus, 2004). In this view, amusia and dyslexia could be entirely distinct disorders, each with its own cognitive cause. The proximity of the brain regions involved (in particular, the superior temporal and inferior frontal cortex, Peretz, 2016; Ramus et al., 2017) would increase the likelihood to be affected together. In our study, five of the 13 children with comorbid amusia and dyslexia had previously been diagnosed with ADD and/or dyscalculia. This could be the consequence of such general susceptibility mechanisms. Yet, this hypothesis remains conjectural and our study does not provide any tangible evidence in its favour or against it. A comparative investigation of the two disorders at the genetic or neural level would be more appropriate to draw conclusions about their biological relationships.

4.3. Cognitive profiles of comorbid cases

While a growing body of research is devoted to the study of the adult form of congenital amusia, little is known about its early characteristics. Indeed, very few studies have been conducted on children, which is probably due to the difficulty of recruiting them given that they are not detected by the school system.

Our results confirm earlier reports (Lebrun et al., 2012; Mignault Goulet et al., 2012) according to which amusia can be observed in childhood and manifests itself in a very similar way to adult cases. The amusic children of the present sample exhibited poor musical abilities despite no history of hearing loss, normal IQ and regular exposure to music. With regard to the last point, the slight difference in exposure to music between dyslexic children and controls can be interpreted in several ways: (i) it may suggest that children with comorbid amusia and dyslexia, who represent more than one third of the sample, are more likely to avoid musical activities because of their musical disorder (ii) it may reveal that their musical environment is impoverished due to family aggregation (iii) alternatively, it may merely be due to the fact that dyslexic children generally have less time to devote to extra-curricular activities.

The musical disorder of children with comorbid amusia and dyslexia was mainly characterized by difficulties in detecting pitch changes in melodies, with about half of them also exhibiting rhythm processing troubles. Whether the latter are independent or consecutive to the pitch deficit would deserve further investigation, as has been done in adults (Lagrois and Peretz, 2019). In their study, Lagrois and Peretz (2019) showed that the beat finding deficit experienced by pitch-deaf adults remains severe whether or not the musical stimulus contains pitch cues. Here, we found that slightly fewer comorbid cases were impaired in the time change detection task ($n = 7$) than in the MBEMA rhythm test ($n = 8$), which supports a partial influence of the melodic context. Further research will be needed to determine whether this discrepancy is due to the nature of the task (beat finding task versus detection of temporal irregularity) or to the stage of development of the participants. Besides, although a majority of comorbid cases performed in the low range in the pitch change detection task, some were unimpaired. This converges towards the hypothesis that amusia is not merely due to a fine-grained pitch discrimination impairment, but may result from higher-level deficits, such as in short-term memory for pitch (Tillmann et al., 2016b).

In addition, most of the comorbid cases showed poor singing performance, especially with regard to the pitch dimension. Their production was characterized by strong pitch deviance and contour errors, as already reported in the adult form (e.g., Dalla Bella et al., 2009; Tremblay-Champoux et al., 2010). Dalla Bella et al. (2009) observed that amusic individuals were even more severely impaired when asked to sing a tune without lyrics. We did not replicate this finding, observing the opposite result pattern: singing turned out to be less successful with than without lyrics. One might attribute this difference to the comorbidity with dyslexia, in which case the verbal condition might require more resources for children with a phonological disorder. However, the fact that the control group obtained a similar pattern of performance is not consistent with this hypothesis. Previous findings have suggested that in the first steps of learning a song, melody and lyrics are remembered separately, making singing a dual task (Racette and Peretz, 2007). In that sense, the difference with former studies on singing proficiency in congenital amusia may be due to the fact that participants of the present study were tested at an earlier stage of development. Another interpretation might be that confidence in the ability to sing is correlated with these variables.

Most interestingly, six of the comorbid cases could sing in tune in spite of their poor pitch perception abilities. This partial dissociation between perception and production has already been observed before, both in congenital amusics (Dalla Bella et al., 2009; Loui et al., 2008; Williamson et al., 2012) and in poor singers (Bradshaw and McHenry, 2005; Dalla Bella et al., 2007; Pfordresher and Brown, 2007). It was further supported by the study of a brain damaged patient who demonstrated selective impairment in singing while his recognition and discrimination of music was preserved (Schön et al., 2004). These findings have sometimes been taken as evidence for functionally distinct auditory pathways for perception and action, by analogy with the dual-route architecture of the visual system (Griffiths, 2008; Loui et al., 2008). Our results provide partial support for this hypothesis. They could also be taken as an argument for some implicit pitch processing in amusic children.

In terms of literacy skills, children with comorbid amusia and dyslexia had impairments comparable to their non-amusic dyslexic peers, all scoring less than −1.5 SD relative to the control group in the literacy accuracy component. This is likely a consequence of the inclusion criteria we used to select participants. Moreover, a phonological deficit was observed in most of them. This was manifested mainly by low phonological awareness/slow access to mental lexicon and, to a lesser extent, by poor short-term verbal memory. As already commented on, the scores on the phonological awareness/RAN component were particularly low in comorbid children compared with those of the other dyslexic children, who did not meet the diagnostic criteria for congenital amusia. In contrast, the verbal memory component appeared to be less discriminating, with few children performing below the −1.5 threshold (see supplementary material: Fig. s2). However, these observations are dependent on how the components were constructed. On the one hand, it cannot be excluded that the digit span tasks, which account for two thirds of the verbal short-term memory component, mask an effect of the pseudoword repetition task, which more specifically assesses phonological memory (and which was, moreover, much more discriminating between the dyslexic and control groups than the two span tasks). In fact, only a moderate correlation was observed between the forward digit span task and the pseudoword repetition task, with the backward digit span task being significantly correlated with neither of these tasks (see supplementary material: Table s1). On the other hand, we only considered accuracy and not response time when computing the phonological awareness/RAN component and the verbal memory component, whereas speed is also sometimes used to characterize the phonological deficit (e.g., Saksida et al., 2016). Indeed, we estimated that response times were not exploitable for these components because they depended on the reaction time of the experimenter, who encoded the child's response on a trial-by-trial basis. These choices may also account for the fact that a high number of dyslexic children performed above the deviation threshold on these two components.

4.4. Potential limitations

The conclusions that can be drawn from the present study are further limited by several other shortcomings. First, our sample size, although twice the size of the seminal adult study (Couvignou et al., 2019), is not large enough to allow a reliable estimate of the prevalence of congenital amusia in dyslexia. Two previous studies (Couvignou et al., 2019; Peretz and Vuvan, 2017), conducted on a larger set of adult participants (266 and 16 625, respectively), suggested that amusia and dyslexia appeared in relative isolation from each other, with a 5.3% amusia rate in dyslexia and a 7.7% dyslexia rate in amusia. However, these findings were limited by the fact that the presence of dyslexia was estimated solely on the basis of self-assessment, without an objective measure of reading performance. Our study suggests that the association between the two disorders may have been underestimated. Still, it needs to be replicated on a larger sample.

Second, dyslexic children were matched to a control group of the same chronological age, but not of the same reading level. Yet, the reduction in reading experience that is inherent in being dyslexic can itself cause differences in sensory processing between participants with dyslexia and controls (e.g., Goswami, 2015; Huettig et al., 2018). Adopting a reading level-matched design would have allowed to control to some extent for the effects of reading experience. Nonetheless, this limitation applies to the conclusions we can draw about group differences but should not have influenced the rate of amusia in the sample.

In the same vein, children with dyslexia scored slightly lower than controls in terms of non-verbal intelligence and exposure to music. The effect of these variables was controlled for in the correlation analyses and they were entered as predictors in the regression analyses. However, they may have exacerbated group differences or artificially increased the deviance thresholds, since these are calculated relatively to the performance of the control group. Nevertheless, these two groups were matched on a variety of demographic and cognitive variables (age, socio-economic level, music education, audio-visual attentional skills and oral language skills) which are rarely considered jointly in studies on developmental or congenital troubles.

Finally, it is worth reminding that the young participants of the present study were French native speakers. One might wonder whether their results can be generalized to other languages. In particular, there has been a growing interest in recent years in the manifestations of congenital amusia in tone languages speakers. About half of those cases showed impairments in the discrimination and identification of lexical tones despite normal production, a pattern which has sometimes been referred to as *lexical tone agnosia* (Nan et al., 2010; Tang et al., 2018). This opens the possibility that lexical tone processing difficulties have an impact on reading acquisition, with stronger links between musical and reading deficits among these speakers than in speakers of non-tone languages. The observations of Nan and colleagues that amusic participants with lexical tone agnosia had normal word and pseudoword reading scores do not support this hypothesis.

4.5. Concluding remarks and implications

Overall, our results suggest that dyslexia and amusia co-occur in a significant proportion of children. Further research will be needed to determine the origin of this co-occurrence and to improve our current understanding of the interaction between the two disorders. In the meantime, these findings have at least two implications for future research and practice.

First, the presence of cases of congenital amusia in samples of dyslexic participants could partly explain the high variability in psychoacoustic performance that has been reported in this population (e.g., Banai and Ahissar, 2005; Banai and Ahissar, 2004). In particular, tasks involving frequency discrimination, pitch memory or, more broadly, musical skills are expected to be less successful in individuals with comorbidity. Therefore, it would be recommended to systematically screen for congenital amusia in experiments involving such skills. Conversely, the presence of developmental dyslexia should be considered in studies assessing language-related skills of amusic individuals, in particular phonological processing skills. This applies whenever one wants to draw specific conclusions about any of the disorders.

Comorbidity should also be taken into account when considering music as a potential adjunct (alongside explicit teaching of reading and speech therapy) in the remediation of dyslexia. Indeed, the characterization of congenital amusia as lifelong disorder suggests that it is difficult to remediate. Training studies in amusia are scarce and have led to mixed results (Anderson et al., 2012; Peretz et al., 2012; Whiteford and Oxenham, 2018; Wilbiks et al., 2016); still, amusic individuals are expected to face greater difficulties than their non-amusic peers in music-based interventions. Besides, according to the OPERA hypothesis (Patel, 2011), one of the five conditions that must be met in order for musical training to benefit the neural encoding of speech is that music activities elicit strong positive emotions. It is far from granted that this condition is met in amusic individuals. Indeed, although they maintain some abilities to perceive emotions from music (Gosselin et al., 2015; Lévêque et al., 2018), their receptivity to music varies: some are fond of it, most of them are indifferent to it, others find it unpleasant and disturbing (Omigie et al., 2012). For the latter, music does not seem an appropriate medium to work with. In any case, amusic cases certainly needs to be handled in a special way, potentially through more intense and longer-term programs.

Credit author statement

Manon Couvignou: Conceptualization, Methodology, Software, Formal analysis, Investigation, Resources, Data curation, Writing – Original Draft, Visualization, Project administration, Funding acquisition. Régine Kolinsky: Conceptualization, Methodology, Writing – Review & Editing, Supervision, Funding acquisition.

Declaration of competing interest

The authors declare no competing interests.

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Appendix A. Supplementary data

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