Absolute Pitch in Autism: A Case Study

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Abstract

We present the case study of QC, a low-functioning adolescent with autism, who possesses the special ability of absolute pitch. QC participated in an in-depth assessment of pitch perception and processing systems relevant for current cognitive models of autism. Her performance was compared to that of mental age- and chronological agematched groups of persons with average intelligence, or to that of musicians. Absolute pitch in identification and production was confirmed. No abnormalities were found in perception of hierarchical (local–global) properties for visual patterns and music. However, a deficit in two components of executive functions, cognitive flexibility and planning for different materials, was evident. Short- and long-term memory for verbal, non-verbal and musical material were without particularities, although QC revealed an exceptional long-term memory for musical pieces when the piano was used for recall. This case suggests that absolute pitch in autism may not result from a multimodal deficit in processing global information. Rather, it may result from a lack of cognitive flexibility in a person with a marked interest for auditory stimuli that occurred at the critical age for the appearance of absolute pitch.

Introduction

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The term 'special abilities' refers to outstanding performances on cognitive tasks, such as calendrical calculation, detection of prime numbers, mental computation, dates and list memory, three-dimensional (3D) drawing, and music memory and interpretation, performed by persons with pervasive developmental disorders (PDD) or, in exceptional cases, with mental retardation only. Their performance in such abilities tends to exceed what would be expected considering their general level of intelligence. Although persons with special abilities are frequently labelled 'savants' (derived from 'savant syndrome'), they do not constitute an independent clinical group per se. However, special abilities are more commonly found in the general context of a developmental disorder of the PDD type. As such, special abilities in persons with disorders in the communication and social area are now incorporated in the 'restricted interest and repetitive behaviours' area of PDD symptoms in the standardized diagnostic scales currently used in research and clinical practice (Lord et al., 1994).

Interest in this area of atypical behaviours has increased in the last two decades following theoretical attempts to characterize the cognitive deficit(s) responsible for PDD symptomatology. Since autism and special abilities often cooccur in one individual, models of deficient behaviours and performances in autism should also be able to account for such special abilities. Nevertheless, interpretation of special abilities is unclear. Some (Ericsson and Faivre, 1988) view special abilities as a by-product of training resulting from a restricted interest for a domain of information. Others (Frith and Happé, 1994) consider special abilities as essential to autism and directly dependent on the cognitive deficits responsible for a large part of the autistic symptomatology. A major difficulty with the interpretation of these phenomena is the apparent lack of common features between the domains of information (music, numbers, words, spatial representations) and the heterogeneity of the cognitive operations performed on these domains (labelling, memorization, computation). However, domains of special ability are all highly structured (Mottron et al., 1998), suggesting that their choice is related to some type of facilitation by this regularity. In addition, striking epidemiological and descriptive similarities are found between persons exhibiting these competencies across the world. These commonalities imply that knowledge of special abilities is essential to a better understanding of PDD and the nature of the autistic disorder.

The heterogeneity of savant abilities may be reduced by classifying them according to (1) the level of complexity of the operations and (2) the coded versus non-coded aspects of the material involved. The first group of special abilities is comprised of perceptual skills that involve the capacity to associate a non-coded element with a coded one, i.e. a tone

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and its name or a visual discontinuity and a drawing feature. The capacity to label or reproduce quickly and directly elements which cannot be labelled or reproduced directly, or at the same speed, by typically developing persons, is also part of this category. One of these rare capacities is subitizing. which refers to the ability to give the numerosity of displayed objects or dots without counting them. Anecdotal accounts of this special ability in autism have been reported; however, further empirical confirmations are required. Another capacity is absolute pitch (AP), the ability to name or to produce pitches without referring to an external standard (Takeuchi and Hulse, 1993). AP has been seen in typically developing persons; however, it may be more commonly detected in persons with autism and savant musical abilities (Young and Nettlebeck, 1995). Miller (1989) suggested that all five cases of musical savants with visual and intellectual impairments that he studied, some of which would be diagnosed with autism according to current criteria, also had AP in production. Nevertheless, diagnostic uncertainty and the confounding role of visual impairments and brain lesions preclude a direct link between AP and persons with autism. A third capacity is the ability to reproduce 3D structures graphically, an ability that is typically only achieved by trial and error by professional draughtsmen (Mottron and Belleville, 1995).

A second group of special abilities is memory skills. This category includes the capacity to memorize the association of several coded elements, digits or words, such as giving somebody's date of birth, knowing the day corresponding to a memorized date, or being able to recognize a memorized prime number as such. Prodigious memory for proper names, telephone numbers and songs are made up of lists of symbolic or coded units (Mottron *et al.*, 1996; Mottron *et al.*, 1999c). These special abilities may be reduced to the memorization of associations between coded items.

A third group of special abilities is operative skills. These skills involve exceptional performance on tasks that cannot be reduced to labelling, reproduction or memorized associations, but which imply some complex operation on coded data (i.e. words, digits or drawn features). These tasks include mental rotations, completion of 3D structural reduction, recognition of unknown prime numbers, and dates or operations of transformation on coded material. This subgroup of operations cannot be reduced to exceptional memory, even if they may rely on, or produce, a high-level memory performance.

According to this tentative classification, AP is an especially informative special ability due to the relative simplicity of the operation involved (pitch labelling) and the early developmental stage at which a similar ability may be found in a typically functioning person. This suggests a causal relationship between AP and autism. AP in reception is found in one of every 10 000 people in the general population (Takeuchi and Hulse, 1993) and is exceptional even in musicians. AP in production is even less common (the exact incidence is not known). Such a rarity, combined with the scarcity of autism, renders the fortuitous association between the two highly unlikely.

Generally considered a non-trainable ability in adults even in contexts where the role of training is emphasized (Ericsson and Faivre, 1988), AP is associated with early musical experience, although the causal relationship between AP and musical training is ambiguous. It is clear, however, that early musical training is not a sufficient condition for the acquisition of AP as most children with early musical training do not develop this ability. When present, AP either precedes precocious musical training, or may be enhanced by musical training if present at a very early age. This distinguishes AP from more complex special abilities like calendrical calculation. In the latter, memory training (for the memory type) or practice of idiosyncratic algorithms (for the operative type) cannot be clearly discarded.

Another interest in the presence of AP in autism emerges from several empirical findings, which suggest that nonsavant persons with autism perform better than comparison participants in memorizing pitch. Savant persons with AP might therefore represent the extreme of an above-average perception of pitch in autism. The superior ability to associate pitch and symbolic material in non-savant children with autism has recently been reported by Heaton et al. (1998). In their study, 10 boys with autism [chronological age (CA): 9.9, mental age (MA): 8.4] and 10 CA-matched comparison boys (CA: 8.1) were presented with stimuli consisting of four tones and four line drawings of animals. In the familiarization session, subjects heard the tone while naming the animals and were told 'this is the animal's favourite sound'. In the pitch identification procedure, children had to point to the drawing corresponding to the presented tone 150 s later as well as 1 week after the learning procedure. Results showed that the group with autism was superior to the group of typically developing boys in their ability to associate tones and pictures in both recall conditions.

Following Frith's (1989) suggestion, Heaton et al. (1998) interpreted their finding as demonstrating 'that an autistic tendency toward segmented information processing leads in turn to stable long-term representations' (p. 301) and that persons with autism possess a 'privileged access to aspects of the physical word that are normally overridden by tendencies toward holistic perception' (p. 302). Frith and colleagues (Frith, 1989; Frith and Happé, 1994) have proposed that savant syndrome abilities may be explained by a non-modular abnormality in information processing. These authors suggest that autism might be characterized by a weak 'central coherence', i.e. a deficit in the tendency exhibited by typically developing persons to integrate information from a given level into more general information. This atypical tendency to focus on the stimulus rather than its context may exist for different material (e.g. auditory, verbal, abstract) and could be applied, mutatis mutandis, at various levels of processing (e.g. perceptual, mnesic). Consistent with this suggestion are findings of superior performance in persons with autism on embedded figures tasks and the Block Design subtest of the Wechsler Intelligence Scale for Children III (WISC-III) (Shah and Frith, 1983, 1993), and of locally oriented graphic construction in EC, a savant autistic draughtsman (Mottron and Belleville, 1993, 1995). We and others (Frith and Happé, 1994; Heaton *et al.*, 1998) saw this as evidence for a locally oriented processing in autism. However, the perceptual interpretation of these particularities has become a matter of debate, as these findings could not be generalized to the general population of persons with autism using purely perceptual tasks (Mottron *et al.*, 1999a, but see Happé, 1996) even though abnormal hierarchization was established in visuo-spatial graphic tasks with an executive component (Mottron *et al.*, 1999b).

The purpose of this paper is 3-fold: (1) to document a case of AP in a person with autism; (2) to characterize her profile of performance with an extensive neuropsychological assessment focusing on relevant areas for cognitive models of autism, such as hierarchical visual and musical perception, executive functions, working and long-term memory, and general visual perception; (3) to discuss how AP and cognitive profile may be accounted for by non-modular models of abnormal hierarchization of information and executive deficits.

Participant

QC was assessed between January 1997 and August 1998, when she was between the ages of 17 and 18 years.

Developmental and medical history

Pregnancy and delivery were normal (6 lb 7 oz, apgar 8,9). Two days after birth, a Streptococcus epidemic occurred in the hospital. She developed a high fever, together with a hyperbilirubinaemia under current critical levels (Day 3 = 12.13 mg/l, Day 4 = 18.31 mg/l, Day 5 = 9.4 mg/l). This was determined to be a meningitis or a septicaemia. Abnormal crying duration and feeding problems were present from birth. QC's developmental milestones were not delayed. She sat at 6 months, walked at 12 months, and was toilet trained for bowel movements at 18 months and urination at 38-40 months. Language appeared delayed (first words at 30 months, first two-word sentences at 48 months). Several yearly episodes of otitis occurred between 1 and 7 years of age. A neuroophthalmological examination at 17 years revealed only abnormally slow saccades. She wears glasses for nearsightedness. Brain magnetic resonance imaging at this age did not reveal any abnormal brain structure.

Genealogy

QC is an only child as her mother had a miscarriage before QC was born. QC's father is colour blind and suffers from stuttering. Colour blindness is also present in three members of her father's family. Her maternal great-grandparents were 15

Table 1a. QC's diagnostic criteria according to ADI-R (Lord et al., 1994) scores

	Age (years)		
ICD-10 criteria		18	Algorithm ^b
Social Interaction			
Failure to use non-verbal behaviours to regulate social interaction (/6)	6		
Failure to develop peer relationships (/10)	8	1	7
Lack of shared enjoyment (/6)	6	5	5
Lack of socioemotional reciprocity (/10)	6	2	6
Total (cut-off = 10) ^a	26	10	23
Communication			
Delay in spoken language and failure to compensate through gesture (/8)	6	6	6
Lack of varied spontaneous make-believe or social initiative play (6)	4	0	
Relative failure to initiate or sustain conversational interchange (/4)	4	2	
Stereotyped, repetitive or idiosyncratic speech (/8)	6	4	5
Total (cut-off = 8)	20	12	16
Repetitive Behaviours and Stereotyped Patterns			
Encompassing preoccupation or circumscribed pattern of interest (/4)	3	4	
Compulsive adherences to non-functional routines or rituals (/4)			
Stereotyped and repetitive motor mannerisms (/2)	1	0	1
Preoccupations with part of objects (/2)	2	2	2
Total (cut-off $= 3$)	7	8	7

Table 1b. QC's current ADOS-G scores

Social scale + Communication Total (cut-off = 10)^a Repetitive Behaviours and Stereotyped Patterns

Total (cut-off = 2)^c

^aValue above which an area is considered sufficiently impaired for the diagnosis of autism.

^bComposite score for the 4-5 years and the current period.

^cThis score is not indispensable for diagnosis.

first cousins. QC's mother presents with a congenital cardiopathy. A case of schizophrenia was reported in QC's maternal uncle, and a first cousin of QC presents with a profile consistent with a diagnosis of autistic disorder with mental retardation. QC's parents are also first cousins.

Psychiatric diagnosis

A formal diagnosis of autistic disorder was performed at the age of 18 years, by retrospective interview with her family regarding her developmental course using the Autism Diagnosis Interview-revised (ADI-R; Lord *et al.*, 1994; Table 1a) and a semi-structured assessment with the Autism Diagnosis Observation Schedule (Lord *et al.*, 1998; Table 1b). QC's early developmental history is highly consistent with autistic disorder with moderate mental retardation. Her first symptoms appeared at 27 months, characterized by a lack of social response (i.e. absence of social play and social withdrawal). When she was 4–5 years of age, her main symptoms were

absence of spontaneous peer-oriented behaviours, echolalia with preservation of original intonation, stereotyped questions, use of an adult's hand as an instrument, delayed echolalia of commercials triggered by hearing part of the repeated sentence, non-functional play (lining cars) and manual stereotypies during happy emotional states. Her main behavioural atypicalities currently consist of an absence of personal social relationships, repetitive and socially irrelevant questions about dates of births, car statistics and car keys, ritualized touching of furniture in unknown areas, ritualized eating habits and a persistent interruption of her daily activity due to her restricted interest for music. One other special ability observed was her capacity to orient herself in places to which she was exposed just once.

History of behaviours related to auditory stimuli and music

A hypersensitivity to noise has been present since childhood. QC tended to scream at the sounds of vacuum cleaners and chainsaws, and would ask questions about the possible presence of these tools in her environment so that she might avoid them. A hearing test at 13 years of age revealed that she was able to detect auditory stimuli 15 dB under the norm for sounds between 1500 and 5000 Hz (right ear), and 10 dB under the norm for sounds between 1000 and 4000 Hz (left ear). The norm represents the volume under which a mean of 1000 typical adults are unable to hear sound. These symptoms were possibly diminished by desensitization to noise at the age of 13 years. A hearing test at 18 years of age showed a marked reduction of sound hypersensitivity, with only a 10 dB peak at 4000 Hz (right ear). QC still avoids microphones for fear of Larsen effects, and jumps with fright in response to low but unexpected sounds.

There are several professional musicians on both sides of QC's family. At least four paternal and two maternal family members are popular, self-taught musicians; however, QC's parents are not musicians and she was not raised in a musical environment. This suggests an important genetic component in her ability. Her interest in music appeared when she was 2 years old. After hearing music only once, she could reproduce tunes she had just heard on a children's keyboard. She was able to sing before any language development. AP has been suspected since QC was 4 years old, before any formal musical training. A real piano replaced the toy piano at home when she was 4 years old. She received 1 h a week of private lessons by a piano teacher, beginning at the age of 5 years, and since that age she has practised less than 2 h a day. Since the age of 14, she has sung in her school choir. QC has been exposed to classical and popular music since she began piano lessons, and she has read music since the age of 10. Her musical memory in natural settings is remarkable in that she is able to play chords of simple harmonization from popular tunes after less than three trials. QC is now 18 years old and she plays classical, moderately difficult pieces like Debussy's 'Arabesque' by memory,

Table 2. QC's general neuropsychological assessment

Tests	Scaled score	Mental age
Wechsler Adult Intelligence Scale-K	Revised	
Information	5	
Digit Span	5	
Vocabulary	5	
Arithmetic	3	
Comprehension	3	
Similarities	7	
Picture Completion	4	
Picture Arrangement	6	
Block Design	7	
Object Assembly	5	
Digit Symbol	4	
Verbal IQ	. 73	(-1 2/3 SD)
	<5 percentile	
Performance IQ	70	(-2 SD)
	<2 percentile	
Full-scale IQ	71	(-1 2/3 SD)
	<5 percentile	
Standard Progressive Matrices	<5-10 percentile	8.5
Peabody Picture Vocabulary Test	<2 percentile	9.6

although she requires a fair bit of practice to commit them to memory. According to her teacher, she is still experiencing technical and expressive progress, especially in emotional expression.

General neuropsychological assessment

A battery of tests assessing general cognitive functioning was administered. This battery consisted of the Wechsler Adult Intelligence Scale-Revised (WAIS-R) (Wechsler, 1981), the Standard Progressive Matrices (Raven, 1938), the Peabody Picture Vocabulary Test (PPVT) in its French form (Dunn et al., 1993) and the Oldfield Lateralization Questionnaire (Oldfield, 1971). QC's Full-Scale IQ (FSIQ) on the WAIS-R was 71 (see Table 2 for details). Her performance on the WAIS-R was typical of persons with autism in that her relative strength was on the Block Design subtest (7) and her weakness was found on the Comprehension subtest (3). QC's IQ on the standard progressive matrices was between 64 and 68 with a MA of 8.5 years. As measured by the PPVT, her MA was 9.6 years, slightly above her FSIQ. According to the Oldfield Lateralization Questionnaire, she is right-handed with a score of 100%.

The special ability of AP was extensively measured and comprehensive testing was carried out to investigate the cognitive systems that may be involved in this special ability. Hierarchical processing in visual and musical perception, as well as executive functions, were especially emphasized, due to their postulated involvement in AP. The hypothesis of an explanation of AP by non-specific superior performance on tasks of working or long-term memory was explored through assessment of these systems for verbal, non-verbal and musical material.

Assessment of specific cognitive functions in the context of a low level of general intelligence raises some comparison difficulties. Deficient performance in a specific area may always be accounted for by an alternative, non-specific explanation of intelligence level. Not considering this factor may lead one to attribute a specific deficit to a given developmental level (Burack et al., 1998). Therefore, QC's performance was compared to that of persons with average intelligence with the same MA (approximately 9 years). When norms for children were not available, QC's performance was compared to that of persons with average intelligence with the same CA. Given that QC's intelligence level fell 2 SD below the average population, normal performance was approximated according to her intelligence level 2 SD under CA norms. Performance at the level of a CA-matched person of average intelligence implies that QC has performed at a level commensurate with her CA and, therefore, above her MA. Performance around 2 SD below her CA corresponds to her MA. Finally, performance clearly falling below 2 SD of CA-matched controls (or under 1 SD of MA-matched controls) would represent an area of specific deficit. These guidelines are based on two considerations: (1) that typically developing children with an MA/CA of 8-9 years perform 2 SD under typical adults; (2) that the error due to this approximation is of the same magnitude for each task. Norms according to different tasks are summarized in Table 4.

Assessment of absolute pitch

Materials and methods

Two abilities were tested in a single session: identification of single piano tones and singing production of named pitches. Pitch identification. In this task, QC heard a single piano tone and was required to write down the correct symbol corresponding to its pitch in musical notation, using no other reference. Since QC exhibited difficulties in writing the proper musical symbol, but was able to produce the correct name orally, she was retested with oral responses at the end of the session. Two pitch identification tests were administered [similar to those used by Ward and Burns (1982) and Zatorre and Beckett (1989)]. In the first test, 12 tones were chosen from a range of four octaves (from C2 to C6) so that all chromatic pitches were presented, but no pitch was repeated in any one octave. The 12 notes were presented four times in random order, for a total of 48 stimuli. In the second test, the 12 notes of the chromatic scale between middle C3 up to C4, including the latter, were used four times in a random order, for a total of 52 stimuli. The isolated piano tones were computer generated from a Rolland CS-55 synthesizer. The tones were 500 ms long, separated by 2 s of silence. QC was presented the stimuli in free field, and for the first test she was given music paper with piano staves (with the top one in treble clef and the bottom one in bass clef), while for the second she was given music paper with only one staff in

 Table 3. Percentages of correct responses given by QC and by normal AP possessors in three tests of absolute pitch abilities

Response mode	QC		Normal AP possessors	
	Written	Oral	Mean	Range
Pitch identification				
Condition 1	71	100	74.8	27.1-100 ^a
Condition 2	88	89.3	89.5	37.3-100 ^a
Pitch production		86.5		

^aZatorre and Beckett (1989), screening tests 1 and 3. There were 18 selfdeclared AP possessors (aged from 13 to 50 years).

bass clef. QC was instructed to write the note she heard on each trial in musical notation. No feedback was given.

Results

The results are summarized in Table 3. In the first test, OC obtained 34 correct responses out of the 48 stimuli. However, it was apparent that her errors were related to the written code. She never went below or above the staves when writing her responses. Therefore, she missed nearly all the notes belonging to the lower octave (C2), which required the writing of supplementary lines. If we discard those tones, QC obtained a score of 31/36, or 86%. Five of the six errors were due to the omission of an alteration (e.g. D3# written as E4) and thus were one semitone below the target. The remaining error was in a four-semitone distance from the target (i.e. D3# was noted as G). It seems relevant to note that D3# was only identified correctly once out of the four presentations. QC never made any octave errors. However, when retested on this condition and asked to identify heard pitches verbally instead of using musical symbols, QC correctly named every single tone, scoring 100%. In naming the pitches, QC systematically used the most frequent labels, i.e. she labelled D# as E flat and A# as B flat. Despite difficulties probably related to the musical writing system. QC definitely possesses AP. Her performance fell well within the accepted range for persons with AP who do not have mental retardation, as can be seen in Table 3.

QC's writing difficulty became more apparent during the second test. After the first 24 trials, QC was asked not only to write the symbol on the staff, but also to write the verbal name of the pitch above the staff. In two cases, she wrote the correct label, but the incorrect symbol. Even with these writing difficulties, QC succeeded in identifying 46 correct musical symbols out of the 52 stimuli (88% correct) and 25 correct verbal names out of 28. The four errors (excluding the two properly named but improperly located in the staff) systematically involved C3#, which was identified as C flat (two semitones distance) in all four presentations. Again, QC's performance accuracy falls well within the accepted range of persons with AP (see Table 3).

It seems clear that QC did not use relative pitch in the

task. In principle, having noted one tone, all the following tones in the test can be derived by relative pitch. There were no consistent errors in any given direction. Error consistency was limited to two pitches: D3# and G3#. Since these notes were presented randomly in the sequence, they clearly support the notion that QC was using absolute, as opposed to relative pitch, although the internal standards of the AP may be unstable for two particular notes (D# and G#). Despite this, QC demonstrated perfect pitch when allowed to name the pitch rather than to write down the symbol of the pitch she heard. This is consistent with the traditional manner in which QC was taught music, with an emphasis on the oral naming of pitches in reading before focusing on learning the written material.

Pitch production. The labels of the same set of pitches that were utilized in the second test were used once again for production. Rather than being presented with the piano tones, QC was presented with the pitch labels of the 13 chromatic pitches, four times in a random order, and asked to sing the label that was presented. The sharp label followed the pitch label whenever there was an alteration (i.e. the term 'flat' was never used) and she was asked to produce C3 either at the low or high end of the series, thereby causing no difficulties. QC's singing responses were recorded on tape. After much insistence, she agreed to sing in the microphone, but was tense for the remainder of the session. Despite her anxiety, her performance was impressive in that she responded immediately, without hesitation, and seemed to do so effortlessly.

Her taped responses were then each compared, by two musicians, to the corresponding keyboard note. The latter judged 45 out of the 52 produced pitches to be perfectly matched, giving her a hit score of 86.5% correct. Two of the seven missed targets were judged to be out of tune by less than a semitone. The remaining five errors were two semitones below the target pitches. These errors affected the notes D3# and A3#. It is noteworthy that QC did not use these labels when identifying the heard pitches. She rather used E flat and B flat, as observed in QC's oral responses in the first test. This mismatch between her internal pitch labels and the ones used in the test may have contributed to the decrease in accuracy observed on this test. However, in the first test, D# also caused problems when a written response was required, and therefore it may not be uniquely related to the inflexible use of some pitch labels.

Overall, QC exhibits clear evidence for an effect of pitch classes in that she never made any mistakes for white-key pitches, the unaltered pitches, in both identification and production. Her errors always corresponded to black-key pitches, the altered pitches. Black-key pitches include a sharp or flat in the pitch class name, and correspond to the black keys on the piano. White-key pitches do not require a sharp or flat in the pitch class name, and correspond to the white keys of the piano keyboard. Nearly every person with AP shows this effect of pitch class [see Takeuchi and Hulse (1993) for a review], making QC similar to the norm in this domain. This pitch class effect is probably related to frequency of exposure, both in everyday musical experience and in early training. White-key pitches are more frequent in Western music and are learned before the black-key pitches.

It is important to emphasize that AP is not an all-or-none phenomenon. As illustrated in Table 3, self-declared persons with AP vary widely in accuracy of AP identification. Moreover, very few of them can produce pitches absolutely. Since there is no documented case of a person with AP who can produce pitches absolutely and cannot identify them absolutely, AP production ability is considered to be characteristic of high levels of AP. Clearly, QC qualifies as such.

In sum, QC exhibits a high level of AP that is qualitatively similar to that displayed by those rare musicians endowed with high levels of AP. Similarly, this endowment does not seem to prevent her from using relative pitch in musical settings so that she may fully appreciate musical structure, which requires relative pitch rather than AP.

Hierarchical processing

One of the purposes of this case study was to assess whether a multimodal tendency to focus on local aspects of stimuli, or abnormal hierarchical processing, could be related to AP. This had been proposed in the context of the 'central coherence' deficit hypothesis, using an ad hoc concept created to account for a series of clinical particularities and empirical findings in autism (Frith, 1989). 'Central coherence' is a concept open to diverse interpretations and operationalizations. One of these deals with the local-global distinction previously established in visual perception and music perception. The functional autonomy of local and global processing in the visual modality has been established through hierarchical stimuli (Navon, 1977). These are patterns where a structure at the 'global' level is formed from patterns at a smaller, or local, level. Arguments for the functional autonomy of local and global processing in visual perception arise from brain injury studies (Robertson et al., 1988), cognitive evoked response potentials studies (Heinze and Munte, 1993) and (functional) fMRI studies (Martinez et al., 1997). In music perception, the global versus local distinction may be interpreted as contour of a melody versus one pitch component of this melody. Studies from brain-injured patients have shown that right versus left brain lesions differentially affect local and global processing in the same direction for visual (Robertson et al., 1988) and musical information (Peretz, 1990). Local/global aspects of visual and musical perception have been studied in the same brain-injured patient (Lassonde et al., 1999), showing a global impairment in both modalities and suggesting the possibility of a common multimodal mechanism. Nevertheless, local/global distinctions have been established and operationalized differently in vision and in audition, and their validity is independent of the existence of a multimodal local/global distinction. Assessing hierarchical perception in visual and musical

modality was especially relevant for QC, since a special ability for pitch labelling was previously interpreted as resulting from the primacy of local processing in music perception (Frith, 1989; Heaton *et al.*, 1998).

Materials and methods

Visual hierarchical processing. Visual hierarchical processing was assessed by measuring reaction time to the detection of (1) letters at the local and global level of hierarchical stimuli (Hierarchization Task), (2) letters of different size (Spatial Frequency Task), (3) embedded versus isolated letters (Inclusion Task) and (4) segmented versus plain letters (Segmentation Task).

In the Hierarchization Task, the stimuli were similar to those used by Lamb et al., (1990) and consisted of computergenerated block letter global patterns of H, S, A and E formed from smaller local patterns of the same letters. The global letters were made up of local letters within a rectangular 5×5 matrix. Three different sizes of stimuli were used and corresponded to three visual angles based on a distance of 1 m from the participant's eves to the computer monitor. The letters H and S were the target letters, and the letters A and E were the distracter letters. Each stimulus presentation consisted of one target and one distracter letter for a total of four stimuli conditions: global H local A, global S local E, global A local H, global E local S. These presentations reflect the factorial combinations of the three variables: level of processing (local or global), target letter (H or S) and distracter (A or E). In the Spatial Frequency Task, the stimuli consisted of computer-generated block letter patterns of large and small letters H and S. In each condition, one of the four possible stimuli was presented alone in the middle of the screen. In the Inclusion Task, the stimuli were letters H or S that were presented either alone, in the middle of the screen, or within a pattern of the same letter (e.g. all H) that formed a 'digital 8'. There were four conditions: H alone, S alone, H in pattern and S in pattern. In the Segmentation Task, the stimuli were letters H and S that were either presented in typical line format or in segmented filled rectangles. There were four conditions: line H, line S, segmented H and segmented S.

In these four tasks, the stimuli were presented in one block of 80 trials, with 20 trials of each of the four conditions. The stimuli were presented for 200 ms with interstimulus intervals that varied randomly from 2200 to 2500 ms. The stimuli were presented with the Instep program on a SVGA monitor connected to a Pentium 100 computer with two response buttons, in a room where illumination was controlled.

Musical hierarchical processing. This task required a 'samedifferent' classification judgement on pairs of melodies (see Peretz, 1987). The melodies were monodic, unfamiliar and tonal in that they started and ended on the tonic, and only included notes from the same key. They contained nine notes of equal duration, and were computer generated with the approximation of a piano timbre. Two types of manipulation were applied to each melody. The first manipulation consisted of transposing each melody to a near key, either by raising each pitch by a perfect fifth or by diminishing each pitch by a fourth. The second manipulation consisted of creating a 'different' melody by changing all pitch intervals while keeping it in the same key, and hence keeping the same initial and ending pitch. In half the cases, the pitch directions of changed intervals were inverted so as to create 'contour violated' melodies; in the other half, the contour was kept the same. Two sets of two practice trials and 48 experimental trials were constructed with these melodies. Each trial consisted of a warning signal, followed by a target melody, then a 2 s silent interval, and finally a comparison melody. The first set of 50 trials corresponded to the untransposed version; it was constructed so that half were made of identical melodies and half of different melodies. These were presented in a random order. The second set of 50 trials, the transposed version, was constructed from the untransposed version by replacing each target melody with its transposed transformation. Therefore, on each trial of this latter set, the target melody was either higher or lower than the comparison melody. Subjects were instructed to ignore this difference in pitch level as if they were to judge the identity of a melody sung by a woman and man. In both the transposed and untransposed set, half the different comparison melodies differed globally by the overall contour or locally by the individual pitches (or pitch intervals).

Results

Visual hierarchical processing. QC's reaction times were indistinguishable from those of CA-matched comparison participants as she performed within 1 SD faster than the reference group for all tasks. In conditions where comparison participants detected local and global targets with a statistically non-significant advantage of local targets, QC even presented a global advantage. Similar to the comparison group, QC's reaction time was slower in the embedded condition. In addition, she was affected by low and high spatial frequencies and segmented versus plain compound stimuli in the same manner as the comparison participants. These similarities demonstrate an absence of impairment in visual hierarchical perception.

Musical hierarchical processing. With the untransposed version, QC's ability to differentiate between different melodies with intact contour ('local modifications') or modified contour ('global modifications') was perfect (24/24). She missed 3/24 targets in recognizing identical trials. With the transposed version, QC missed three 'different' trials, which she judged as identical, and three 'identical' trials, which she judged as different. These errors concerned two 'local' and one 'global' difference, hence showing evidence of a normal (i.e. global) contour superiority effect [see Peretz (1987) for a literature review on typically developing persons]. Interestingly, QC's

excellent performance (87.5% correct) with the transposed version shows that she is able to use relative pitch melody discrimination. This is so because, when transposing melodies, all pitch intervals are kept the same, whereas not a single AP is maintained. Therefore, if QC were only relying on AP, she would have judged all transposed melodies as different, something she did not do. This particular aspect of the results shows that QC's AP endowment does not prevent her from using relative pitch in musical settings, permitting her to appreciate musical structure fully, which requires relative pitch rather than AP.

Executive functions

An executive function deficit has been reported in persons with autism [for a review, see Ozonoff (1998)]. Furthermore, a deficit in cognitive flexibility, a component of the executive functions, could explain the symptoms found in restricted areas and in repetitive behaviours in persons with autism (Russell, 1997; Turner, 1997). It is thus pertinent to study the integrity of QC's executive functions. Typical executive tasks, as well as tasks possibly related to executive components, were administered to assess the main components of executive functions at various levels of complexity, and in various modalities.

Materials and methods

Cognitive flexibility. QC's capacity to switch overt attention between representations was assessed with Luria's Graphic Seriation Task which includes alternated sequences of simple patterns (Luria, 1966), as well as a Piagetian AB Task (Piaget, 1954). Efficient performance on this Piagetian Task requires the participant to identify correctly which of two cups has an object hidden under it. QC's capacity to switch covert attention between mental representations was measured by the Trail B Task (Speen and Gaddes, 1969), where the subject has to link alternatively digits 1-13 and letters A-L. At a more abstract level, QC's capacity to switch covert attention between rules was assessed by the Wisconsin Card Sorting Task (Heaton, 1981). This task requires the participant to extract a rule from feedback provided by the examiner, and to switch this rule when the feedback has changed. An adapted version of the Uses of Object Task (Guilford et al., 1978) and the Alternate Uses Test (see Lezak, 1995) were also utilized. In these tests, the participant must give the maximum number of different uses for a given object, including uncommon uses. This provides a qualitative assessment of convergent versus divergent thinking, cognitive flexibility and initiation.

To ensure that QC's difficulties with cognitive flexibility were not due to general cognitive impairment, the Spatial Reversal Task (Kaufman *et al.*, 1989) was used. This task is a simplified version of the Wisconsin Card Sorting Task and was devised for non-human primates and very young children, both clearly falling within QC's general level of cognitive functioning. In this task, the subject is required to find the reward hidden under a cup without seeing the experimenter placing it. Every four trials, the cup under which the reward is placed changes. The participant must recognize and learn this simple rule, and modify his/her answer according to task demands.

Planning. An attempt to use the Tower of London with QC failed, as the sequence of instructions was above QC's level of understanding. Planning was therefore assessed through copying of Rey's complex figure (Osterrieth, 1944) and the Maze subtest of the WISC-III (Wechsler, 1991).

Inhibition and resistance to interference. QC's ability to filter extraneous stimuli was deficient in natural settings. QC was easily distracted by both internal stimuli, such as repetitive questions triggered by elements in the environment, and external stimuli, e.g. the irrelevant stimuli present in the Raven's task or incidental noises heard in the room. We used an auditory continuous performance task to measure resistance to interference in the auditory modality. In this task, the participant is required to identify 25 targets (the letter A) presented among distractors (other letters). The participant must raise his/her hand when he/she has heard the letter 'A'. Task duration is 5 min and stimuli are presented on a tape recorder. Resistance to interference in the visual modality was measured by the Boston Cancellation Task (Weintraub and Mesulam, 1988) and the Stroop Task (Golden, 1978). The Boston Cancellation Task requires the participant to cross out letters randomly scattered among distractors. The Stroop Task requires the participant to read 100 printed names of colours as quickly as possible in the baseline 'word' condition. In the 'colour' condition, the participant must say the name of the colour of the presented stimuli, and in the 'interference' condition, he/she must read the names of colours that are printed in a different colour. Inhibition is indexed by misreading of colour names and resistance to interference by the difference between baseline and interference condition.

Initiation. The capacity to initiate was assessed in four ways. First, with the Verbal Fluency Task, a task that requires the participant to provide as many words as possible within a given category. Second, with the execution speed subtest of the Boston Cancellation Task. Third, with the baseline execution speed in the Stroop Task, and finally with the Trail A (Speen and Gaddes, 1969), where participants must sequentially link the digits 1–25.

Indirect evaluation of executive functions. 'Mentalizing' ability was assessed through a French translation of Happé's (1994) 'short stories'. These stories emphasize irony, the figurative use of language, and metaphors and humour. As such, they require 'mentalizing', an ability that is impaired in low-functioning persons with autism. Typical executive tasks as well as tasks possibly related to executive components, such as Theory of Mind tasks, were administered to assess the main components of executive functions at various levels of complexity and in various modalities. Working memory tasks with an executive component are presented in the Working memory section.

Results

Cognitive flexibility. QC was very slow in alternating between letters and digits in the Trail B (inferior to 11 SD), as well as in alternating abstract rules in the Wisconsin Card Sorting Task, where she performed under 5 SD for perseverations. In the Luria Graphic Seriation Task, she also presented numerous graphic perseverations. QC was able to provide various correct, but stereotyped, uses for a given object, but could not furnish an uncommon use in the Uses of Object Task/Alternate Uses Test. In the Spatial Reversal Task, QC was unable to find the alternative rule, and presented loss of sequence as well as perseverative errors. In contrast, her performance on the Piagetian AB Task was not impaired. This suggests that the planning and/or flexibility component is responsible for her low performance in the Spatial Reversal Task, rather than a delay in mastering object permanence. According to Ozonoff (1998), the detection of a lack of cognitive flexibility may be more likely through behavioural observations of perseverative behaviours than with formal testing. OC's repetitive questions and topic perseverations presented as some of her most problematic and socially inappropriate behaviours in natural settings.

Planning. Graphic planning was QC's most impaired planning ability as she performed under 3 SD below CA norms in copying the Rey complex figure, with several displaced or missing elements. In contrast, on the Maze subtest of the WISC-III, a task assessing spatial planning, QC performed between her MA and CA, indicating that her spatial planning ability is better developed than her graphic planning ability. A possible explanation for this dissociation is that impaired planning, in general, may be compensated for by intact or overdeveloped spatial abilities. This seems to be a valid explanation, as it is consistent with QC's remarkable spatial orientation and recognition abilities.

Inhibition and resistance to interference. QC did not miss any targets or exhibit any fatigue effect in the Auditory Continuous Performance Task. Similarly, within the visual modality, QC was not impulsive and did not commit any commission errors in the Boston Cancellation Task. In the classical Stroop Task, QC did not appear sensitive to interference between colour and names of colours, as she performed slightly above norms for reading speed. It is worth noting that performance on the Stroop Task is generally intact among persons with autism (Bryson, 1983; Eskes *et al.*, 1990).

Initiation. Verbal fluency was within normal limits; however,

QC's execution speed was generally very slow (Stroop: <5 SD; Trail A: <8 SD). This finding cannot be attributed to a general lethargy related to low intelligence given that QC's performance was even faster than CA-matched comparison participants on detecting letters. As the above-mentioned tasks require less initiation by the participant, this finding suggests that QC's initiation component is impaired.

Indirect evaluation of executive functions. QC failed all the 'short stories' as she was unable to understand pretending, irony and deception, all of which are hypothesized to rely heavily on an executive component (Russell, 1997), although failure may be related to an impairment in mentalizing abilities (Happé, 1994). For example, QC interpreted the French metaphorical expression 'avoir un chat dans la gorge' (to have a cat in one's throat), which refers to feeling the need to cough, literally as opposed to metaphorically.

Memory

Memory assessment was performed in order to establish whether general properties of QC's memory could explain her special ability in music.

Working memory

Materials and methods. The phonological loop, visuo-spatial scratch pad and central executive of working memory were assessed (Baddeley, 1986). QC's working memory for auditory non-verbal material was subsequently tested.

(i) Phonological loop. The phonological loop was measured with a syllable, digit and word span, and with measurement of the word length and phonological similarity effects. The span tasks required the immediate serial recall of verbal sequences of increasing length, beginning with two-item sequences. Nonsense syllables were used as a verbal counterpart of the non-verbal auditory span described below (S.Belleville, in preparation). Only two syllables (RAN-BIJ) were used and they were repeated in a series. The word length effect was verified by comparing immediate recall of short versus long words (Duchesne et al., 1998). The phonological similarity effect was measured by comparing immediate recall of rhyming versus non-rhyming words. Items were presented in the auditory modality and response was oral. Participants were tested with sequences whose length corresponded to the digit span capacity of the participants. There were 10 sequences in each condition.

(ii) Visuo-spatial scratch pad. Immediate memory for visuo-spatial material was assessed through face and location span tasks (Duchesne *et al.*, 1996). In the face span task, series of photographs of unfamiliar faces were presented sequentially in the middle of a computer monitor. Immediately after the presentation, a 2×4 face matrix was shown. Participants were asked to point to the faces that were just presented in their correct order. As in the usual span procedure, sequences were presented in an increasing order with

Table 4. Summary of QC's performances, compared to mental age (MA)-matched and chronological age (CA)-matched control groups

Task 	QC	Control group (SD or range)	Type of control group
Hierarchical processing Vision			
Hierarchical stimuli			
Callebel targets	547 ms	525 (117)	CA-matched control group (G)
"global targets	503 ms	550 (141)	CA-matched control group (G)
Spatial frequency			
cahigh spatial frequency	530 ms	514 ms (96)	CA-matched control group (G)
^{ca} low spatial frequency	520 ms	523 ms (111)	CA-matched control group (G)
Inclusion task			
caincluded	591 ms	561 ms (96)	CA-matched control group (G)
^{ca} in isolation	556 ms	516 ms (72)	CA-matched control group (G)
Segmentation task			8F(-)
casegmented	459 ms	505 ms (101)	CA-matched control group (G)
^{ca} plain	452 ms	498 ms (98)	CA-matched control group (G)
Music			err matched conder group (G)
Non-transposed condition			
! unmodified	20/24		
! local modification	12/12		
! global modification	12/12		
Transposed condition			
! unmodified	21/24		
! local modification	10/12		
! global modification	11/12		
Executive functions			
Cognitive flexibility			
Trail B	8'45'	27' (12.8)	
Wissensin Cand Section Text	8 45	27 (12.8)	CA-matched published norms (Davies, 1968)
magatagariag	4		*
	4	5.6 (1.0)	CA-matched published norms (Heaton, 1981)
Luria's seriation	Ovalitativa immediane	10.4 (8.0)	CA-matched published norms (Heaton, 1981)
Spatial Reversal Task	Qualitative impairment	-	
Use of Object Test	Qualitative impairment		
Alternative Uses Test	Qualitative impairment		
Diamife estim	Quantative impairment		
Funification	20		
maMara took	30	69 (for scaled score: 10)	CA-matched published norms (Denman, 1987)
Inhibition interformed	8	10 (1.5)	CA-matched published norms (Wechsler, 1991)
caStroop	7 56	0 (10)	
^{ca} Continuous Performance Task	7.50	0 (10)	CA-matched published norms (Golden, 1978)
Boston Cancellation Task	23123		CA-matched unpublished norms (E)
Boston Cancentation Task			CA matched sublided source (TVL's at the tar
caStructured	0 omissions		CA-matched published norms (weintraub and Mesulemy,
Shavaida	0 01113310113	64 (2.22)	(1988)
Non-structured	9 omissions	.04 (2.22)	CA-matched published norms (Weintraub and Mesulemy,
Initiation	> 01115510115		1968)
Trail A	1/07//	146160	
Verbal fluency	10/	14.0 (0.2)	CA-matched published norms (Davies, 1968)
^{ca} Phonemic	0	14 (7 (5 74)	T T 1 11 1
^{ca} Semantic	0 11	14.07 (5.74)	Unpublished norms
The sector of Mark	11	20.07 (0.83)	Unpublished norms
Theories of Mind	0.77		
□ <u>short stories</u>	0%	87% (45–100%)	MA-matched control group, (Happé, 1994)
Working momony			
Verbal material			
maWord span	4	4.55 (0.50)	
maDigit span	4	4.55 (0.52)	CA-matched control group (A)
maSyllable span	4	0.41(0.93)	CA-matched control group (C)
^{ca} Phonological similarity effect	+	3.33 (3-4)	MA-matched control group (B)
rhyming words (corr trials)	5		
non-rhyming words	10		
^{ca} Word length effect	10		
short words (corr trials)	10		
long words	8		
Alpha span	v		
madirect recall	3	(2(17))	
maalphabetical recall	5 1	0.3 (1.7)	CA-matched control group (C)
^{ca} loss	1 66%	5.4 (4.0) 120- (270-)	CA-matched control group (C)
	50 <i>10</i>		CA-matched control group (C)

Task	QC	Control group (SD or range)	Type of control group
Divided Attention Task ^{ca} span loss ^{ca} tracking loss	-1.7	0.9 (1.4) 2.9 (3.6)	CA-matched control group (D)
Visuo-spatial material ^{ma} Face span Spatial location span ^{ma} simultaneous condition	5	3.18 (0.75) 7 (1.18)	CA-matched control group (A) CA-matched control group (A)
^{ma} sequential condition Musical material	3	5.18 (0.98)	CA-matched control group (A)
Musical sequence span	4	4.55 (4-5)	MA-matched control group (B)
Long-term memory Verbal material	12/24	19 66/24 (18 22)	MA matched control group (R)
^{ma} Word recognition Buschke	19/24	22.33/24 (22–23)	MA-matched control group (B)
Free recall (trial 1)	7/16	11.3/16 (1.9)	CA-matched published norms (Grober and Buschke, 1987) CA-matched published norms (Grober and Buschke,
Free recall (trial 2)	9/16	13.5/16 (1.5)	1987) CA-matched published norms (Grober and Buschke
☐ Free recall (trial 3) ^{ca} recognition Semantic Cued Recall	9/16 16/16	14.5/16 (1.3)	1987)
^{ca} trial 1 ^{ca} trial 2 ^{ca} trial 3	7/16 7/16 6/16		
Rey's 15 words ^{ma} Trial 1 ^{ma} Trial 2 ^{ma} Trial 3 ^{ma} Trial 4	2/15 5/15 9/15 10/15	6.17 (1.70) 8.67 (1.97) 10.42 (1.78) 12.08 (1.51)	CA-matched control group (D) CA-matched control group (D) CA-matched control group (D) CA-matched control group (D)
^{ca} Trial 5	11/15	12.66 (1.97)	CA-matched control group (D)
□ Rey figure	inferior to percentile		CA-matched norms (Denman, 1987)
Musical material ^{ma} Musical recognition ! Musical recall (piano)	16/20 0.34	17/20 (16–18) 0.13 (0.13)	MA-matched control group (B) CA-matched, control group (H)
Visual perception ^{ca} Length matching ^{ca} Orientation matching ^{ca} Position of gap matching ^{ca} Minimum feature matching ^{ca} Figures matching ^{ca} Superimposed figures ^{ca} Functional matching ^{ca} Categorical matching ^{ca} Object decision task	28/30 25/30 36/40 24/25 9/10 33/36 10/10 10/10 23/24	26.9 (2230) 24.8 (1829) 35.1 (2439) 21.6 (16.6-25) nl < 3 errors id. id. id. 23/24	Adults published norms (Riddoch and Humphreys, 1993) Adults published cut-offs (Agniel <i>et al.</i> , 1992) Adults published cut-offs (Agniel <i>et al.</i> , 1992) CA-matched control group (F)

□, specific deficit (performance under 2 SD of chronological age-matched control groups or under 1 SD of mental age-matched controls). ^{ca}Normal performance (equivalent to chronological age-matched control group). ^{ma}Performance equivalent to mental age-matched control group. !Peak of ability (performance superior to 1 SD of chronological age-matched control group). Type of reference group used:

Reference groups	CA (means)	Expertise		
(A)	15.7	11	Non-musicians	
(B)	9	3	Non-musicians	
(C)	22.3	16	Non-musicians	
(D)	14.9	12	Non-musicians	
(E)	8 to 16	108	Non-musicians	
(F)	15.6	14	Non-musicians	
(G)	14.8	11	Non-musicians	
(H)	18	3	Musicians	

four trials per sequence length. This started with two-item sequences and ended when the subject failed on half of the series. A similar procedure was used in the spatial localization span task. Sixteen empty squares disposed at random were presented on a 'touch-screen' computer. Some squares blackened for a short period of time, either simultaneously or sequentially, starting with short series of squares and increasing the length of the series until the subject failed on 50% of the trials. In both conditions, the subject had to point to the blackened items immediately after presentation. Whereas order was not a factor in the simultaneous condition, subjects had to report the correct order in the sequential condition.

(iii) Central executive. The executive component of working memory was measured with a task that requires active manipulation of the content of working memory. The alphaspan paradigm (Belleville et al., 1998) compares recall of lists of words in their order of presentation as compared to in alphabetical order. There were 10 trials in each condition. The length of the series to be recalled corresponded to the participant's word span. Divided attention was also measured with a task that requires the participant to perform serial recall and visual tracking concurrently (Belleville et al., 1992; Chatelois et al., 1993). Tracking speed was established before the task in order to obtain a success rate of 70%. Furthermore, the length of the lists to be recalled corresponded to the participant's digit span minus one item. Performance in a condition of divided attention was compared to that in a condition of full attention where both tasks were completed in isolation.

(iv) Auditory non-verbal store. Auditory non-verbal working memory was tested with binary sequences of sounds (low pitch: C4; high pitch: C5; S. Belleville, in preparation). Sequences made of these two notes were presented at the rate of one item per second. Immediate serial recall required the participants to maintain the high and low values of the two notes. As is commonly done, the number of items was gradually increased.

Results. (i) *Phonological loop.* Table 4 shows QC's performance on all working memory tasks. QC's span for digits (4), syllables (4) and words (4) was comparable to that of MA-matched comparison participants. Furthermore, she exhibited clear word length and phonological similarity effects. These results indicate typical functioning of the phonological loop.

(ii) Visuo-spatial scratch pad. QC obtained a face span of two. In the location span, she obtained five in the simultaneous condition and three in the sequential condition. These results are within 2 SD of the mean of CA-matched comparison participants, indicating no specific impairment of this component.

(iii) Central executive. QC's recall was three in the direct condition and one in the alphabetical conditions, with many intrusions in the latter. This corresponds to a loss of 66% (CA comparison participants; X = 43%, s.D. = 37.9) incurred by alphabetical recall [(alphabetical minus direct)/alphabetical]. In contrast, QC's ability to divide her attention between two tasks was typical: when performed in conditions of divided attention, her ability to recall digits decreased in a manner that was similar to the comparison group, and her performance on the tracking task even improved.

(iv) Auditory non-verbal store. QC obtained a non-verbal auditory span of four, identical to that obtained by MA comparison participants. QC's working memory for nonverbal material was not superior to her MA, even when musical material was involved. Furthermore, it is similar to her verbal span (syllable) assessed in similar conditions.

Long-term memory

Materials and methods. (i) Episodic memory for verbal material. This was assessed with a word and non-word recognition task (Belleville et al., 1997) that measures the influence of long-term memory on memorization, a free and cued recall task (Grober and Buschke's Task; Grober and Buschke, 1987) that separates the encoding and retrieval components of long-term memory, and a list-learning task (Rey's Auditory-Verbal Learning Test; Rey, 1964) that investigated QC's learning curve, as well as recency and primacy effect. The word and non-word recognition task required the recognition of 12 disyllabic concrete words of average frequency and 12 disyllabic non-words. Memory for the two types of material was tested separately. Items were first read twice in a different order, and were then presented for recognition with 12 distractors. Half of the distractors were phonologically similar to the targets (according to their first or last syllable). In Grober and Buschke's Task, the free recall phase consisted of a visual presentation of 16 words belonging to different categories. The words were presented in groups of four items. Encoding was oriented toward the semantic properties of words as participants were asked to point to the word belonging to a given category and learning was ensured after each four-word presentation. In the recall phase, participants performed three successive recalls of the learned list. For each trial, non-recalled words were cued by their semantic category. In the recognition phase, the list of 16 words was presented among 16 distractors. The Rey Auditory-Verbal Learning Test consisted of five successive presentations and recalls of a list of 15 semantically unrelated words.

(ii) *Episodic memory for graphic material.* This was assessed by recall of Rey's complex figure (Osterrieth, 1944). The Rey figure consists of a complex geometric design to be copied and reproduced immediately after viewing, and provides a quantified assessment of visuo-spatial memory.

(iii) Long-term memory for musical material. This was assessed with one recognition and one recall task. The melody recognition task (Belleville et al., in preparation) is an incidental learning task that follows the same design as the word and non-word recognition task. Ten unfamiliar, wellformed Western melodies were played twice in a different order. In the recognition phase, these melodies were presented with 10 distractors matched in structure with the target melodies and participants were asked to indicate the melodies that had been presented in the learning phase. In the music recall task, nine piano pieces of different lengths were composed. The duration of the pieces varied from 19 to 76 s and they contained 8-44 measures written in both the treble and bass clefs. Each piece was presented in the auditory modality via a synthesizer. Participants were required to play each piece on a piano keyboard immediately after hearing it. A professional pianist scored QC's performance by measure; +1 indicated that the rendition was accurate and -0.5 indicated error along four dimensions: the right-hand rendition, the left-hand rendition, melody and rhythm, and as a function of the total number of measures in the piece. A composite score was computed by averaging the four scores across the nine pieces. QC's performance was compared to that of three CA- and gender-matched amateur pianists (with 8 years of piano lessons) with average intelligence and no AP.

Results. (i) Verbal material. QC performed at chance level in the non-word recognition task, well below MA-matched comparison participants. In the word recognition task, she also performed under MA-matched comparison participants, but was above chance. In Grober and Buschke's Task, performance in free recall was very poor in all three trials. By contrast, QC was able to recall 91% of missing words with the aid of the semantic cues. Her cued recall score was thus average. Some intrusions of items belonging to the same semantic category were observed. The contrast between impaired free recall and average cued recall suggests an impairment in her ability to organize information retrieval spontaneously. The same dissociation was observed following frontal lesions, and has been attributed to an executive deficit. In the Rey Auditory-Verbal Learning Test, QC performed under 2 SD for trial 1, under 1 SD for trial 2, and within the average range for trials 3-5. That QC was able to perform typically after a few learning trials supports the hypothesis of ineffective or slower organization of encoding and retrieval processes.

(ii) *Graphic material*. Graphic recall of Rey's complex figure was very poor (inferior to the first percentile). Graphic delayed recall was fraught with additions, perseverations and rotations. This finding must be interpreted in relation to her excellent recognition of places in natural settings. QC may present the same contrast between low recall and high recognition for visually presented material as between free and cued recall for words, supporting a deficit in information recovery, but not in information storage.

(iii) *Musical material*. Results obtained in the Melody Recognition Task showed that QC was not exceptional for melody recognition since her performance was similar to MA-matched and not CA-matched comparison participants. Nevertheless, her musical recognition was slightly stronger than her verbal recognition. In contrast, her piano recall of musical material was exceptional. QC performed 1 SD above a comparison group of CA-matched persons with average intelligence with similar musical experience. Incidentally, QC was able to play two stimuli of this task in the original key, with very few errors, over 6 months later.

In summary, the efficiency of QC's working and longterm memory for various types of materials seems to depend on task requirements. On tasks with a planning component (free recall), her performance was clearly impaired. On span, learning and recognition tasks, her abilities were commensurate with her MA, even when the material was musical. On tasks without a planning component (cued recall), her performance was average. Finally, QC's performance was exceptional on tasks requiring the recall of musical material when using the piano as a recall instrument.

Visual perception

There are several relevant reasons to assess QC's visual perception. According to Miller (1989), most musical savants have a visual impairment. Second, her visual explorations are atypical in social interactions in that she will scan the hemispace opposite to the examiner, with the exception of brief saccades in the relevant direction. Finally, the hypothesis of abnormal hierarchical perception in the visual modality predicts atypical object recognition.

Materials and methods

Low-level perceptual processes were assessed with the following subscales of the Birmingham Object Recognition Battery (Riddoch and Humphreys, 1993): the Object Copy Task, the Length Match Task, the Size Match Task, the Orientation Mask Task and the Position of Gap Match Task. High-level perceptual processes, i.e. construction of object representation and object recognition, were measured with three other subscales of the same tool: the Minimal Feature Match Task (matching of objects under two different viewpoints, one viewpoint that masks the objects' main identifying feature), the Matching of Objects Under Canonical and Noncanonical Views Task, and the Object Decision Task with possible and impossible figures. Finally, identification of overlapping figures and functional and categorical matching tasks were borrowed from the Protocole d'évaluation des gnosies visuelles Montréal-Toulouse (Agniel et al., 1992).

Results

Low- and high-level visual perception. QC's performance on the Length Match Task, the Orientation Match Task, the Position of Gap Match Task, The Minimal Feature Match Task and the Object Decision Task was identical to that of adults with average intelligence. QC was equally able to identify superimposed figures, and perform tasks requiring functional and categorical matching.

Discussion

Data summary

QC presents with a clear diagnosis of autistic disorder (Table 1) with low intelligence (Table 2). She possesses a special ability for absolute judgement and production of pitch (Table 3), as well as an exceptional long-term memory for music that she is learning and reproducing on a piano keyboard. An in-depth assessment of hierarchical perception of visual and musical material revealed that her perception of global properties of musical and visual patterns is comparable to persons with average intelligence. Other intact, CA-appropriate abilities include sustained attention, spatial orientation and storage component of long-term memory. On working and long-term memory tasks, her performance is MA appropriate. Using our approximation between MA and low general intelligence, we may conclude that QC's main neuropsychological deficit involves two executive components: planning in free recall and graphic tasks, and cognitive flexibility in graphic tasks, rule finding and discourse (Table 4).

The purpose of the present case study was to investigate which cognitive deficits or particularities are related to the apparition of a special ability, AP. Group studies of persons with savant syndrome typically assess only a limited number of factors, and are consequently more prone to missing relevant cognitive factors. Conversely, in-depth case studies are more exhaustive and therefore are less likely to overlook relevant deficits. The following discussion will begin by focusing on the way in which the presence of AP in QC, an adolescent with autism, may relate to her cognitive profile of performances across a large number of abilities. Following this, the focus will switch to the strength with which cognitive models alone can account for special abilities, i.e. the 'hidden variable' issue.

Which cognitive deficits may be related to QC's absolute pitch?

QC's unique profile presents us with the opportunity to test the veracity of the hypotheses of a local bias (Frith, 1989) or absence of a particular bias (Mottron and Belleville, 1993) in the information processing of persons with autism possessing special abilities in music perception and memory. These hypotheses may be addressed either in the specific domain of the special ability (music perception) or in other domains (e.g. visual perception). In music perception, QC is a person with AP, i.e. who possesses exceptional abilities to perceive and produce pitches. She also processes relative pitches at the level of a musician. If 'local' aspects of musical information are understood as absolute information relative to single pitch, and 'global' aspects as relative tonal information, QC's AP cannot be accounted for by a deficit in global musical processing. Moreover, in visual perception, QC does not present a locally oriented processing bias, either in general object and pattern matching and recognition, or in reaction time tasks directly tapping visual hierarchical processing. In sum, and contrary to the suggestion put forward by Heaton et al. (1998), it appears that a theory based on a multimodal tendency towards locally biased perceptual processing is limited in its ability to account for QC's special ability for pitch identification and production.

Another outcome of the current findings concerns identifying the nature of the relationships between the peaks and deficits in performance for a given participant. Predictions by the 'Central coherence' theory vary in their level of generality and their domain of validity. In a 'within-level' interpretation of this model, persons with autism might favour one channel in a given level of processing, e.g. favour a local versus global channel, within the level of construction of visual representations. This interpretation has found contradictory evidence in non-savant, high-functioning persons with autism (Brian and Bryson, 1996, but see Joliffe and Baron-Cohen, 1997; Mottron et al., 1999b). A 'betweenlevels' interpretation of this model considers the relationship between lower (i.e. data-based and modular) and higher levels of processing. An example of this interpretation is Happé's (1997) account of the low performance of persons with autism on a homograph disambiguation task according to context. Persons with autism made little use of the context of preceding sentences in pronouncing homographs, suggesting that they favoured processing at the level of lexical units versus other levels, i.e. verbal context or semantic processing (in which lexical units are integrated for typically developing subjects). In this scenario, a deficit in higher level processing would be compensated for by placing greater importance on data-based levels. Heaton et al. (1998) suggested that typical persons lose the ability to memorize pitch because pitch perception is normally integrated into more complex levels of processing along development (Trehub and Trainor, 1993), resulting in a loss of absolute properties. Conversely, savant persons with autism maintain this ability due to a defect in processing these superior levels. A similar explanation was put forward by Mottron et al. (1996) in a case of exceptional memory for proper names. That is, a semantic deficit would result in a specific memorization for lexical units to compensate for other defective levels of processing. QC's current profile of performance does not support this interpretation as no deficit in perception of global aspects of musical information is found, nor any overdevelopment of data-based levels in domains outside her special ability, e.g. non-verbal phonological material. In summary, the hypothesis of an imbalance between two complementary levels, local and global, with the overuse of one channel resulting from a deficit in the other, is not confirmed here.

Cognitive flexibility represents the only cognitive operation for which QC clearly performed below her MA. If we assume that the association between this deficit and AP is not due to chance, impaired cognitive flexibility is the most plausible cognitive deficit related to QC's outstanding ability for pitch processing. In this light, an executive deficit should be responsible for a special ability, as suggested by the executive account of repetitive behaviours and restricted interests in autism (Turner, 1997). However, the absence of specificity of executive deficit in persons with autism and a fortiori for persons with savant syndrome negates the plausibility of considering executive deficits as the unique factor responsible for the apparition of AP. Mottron et al. (1999c) propose that executive deficits may be responsible for overtraining in certain types of operations, but not in the choice of the operation.

Are cognitive deficits alone able to account for absolute pitch?

In their account of special abilities, both Central Coherence Hypothesis theory (Frith and Happé 1994) and Executive Deficit Hypothesis (Turner, 1997) emphasize multimodal deficits. These models hypothesize the same deficit for different domains of data, e.g. language, visual patterns, music and memory. Both theories attribute an important part of the clinical picture of autism, as well as the idiosyncratic processing of the domain concerned with the special ability, to the same deficit. However, multimodal models of special abilities are unable to account for the fact that special abilities are most often present in only one modality for a given person. For example, OC exhibits a dramatic discrepancy between her mastery of pitch labelling and production, and her lesser ability to process other domains of information, even within the area of music. In the rare published cases where a participant has been shown to possess several special abilities, none have been shown to have all the known special abilities. In addition, these models cannot explain the restricted number of cases with special abilities in the general population of persons with autism. Taken together, these elements suggest that a 'hidden variable' is required, in addition to autism and cognitive deficits, to possess a special ability.

Cognitive models that attempt to account for special abilities must explain its presence in only a limited number of persons with autism. QC's case illustrates that detection of auditory information under the usual threshold and/or musical giftedness accompanied by a critical period of musical training may be among the possible 'hidden variables' required for development of this special ability. First, QC seemed to possess a particular interest for auditory stimulation at an early age, as suggested by her oversensitivity to sounds. In addition, the absence of music in her environment while growing up, and the high prevalence of musicians in her family, suggests a genetic component for this interest. Second, this innate interest had to be fed by musical training and by the accessibility of an instrument. Third, the strong link between AP and development (Takeuchi and Hulse, 1993) suggests that this 'hidden variable' might act only in a critical period early in development. The probability that a child will possess AP and the accuracy of the AP identification increase as the age at which the child receives musical training decreases. There is also a greater success in training AP in children than in adults, and a shift in the reproduction of melodies from absolute (pitch) to relational aspects of the pitch (direction of change, pitch intervals) during development. In addition, a lack of cognitive flexibility may account for the observation that a domain in which an interest exists is more frequently explored or practised through more repetitive experiences and with less variability in persons with autism that in typically developing children. Finally, the occurrence of this practice during a critical developmental period for pitch memorization might favour the creation of long-term memory units for pitch.

Contrary to the uniqueness of the cognitive deficit hypothesized in various persons with autism, this 'hidden variable' cannot be generalized to all special abilities as it probably changes for each individual. According to the proposed classification for special abilities discussed in the Introduction, perceptual abilities might favour a perceptual hidden variable. Comprehensive case studies of persons with autism presenting with other types of special abilities are necessary to determine if, and what kind of, 'hidden variables' are required for memory and operative special abilities.

What does QC teach us about music perception in non-savant persons with autism?

Our findings suggest that QC's superior performance in pitch perception may result from the combination of a low-level oversensitivity to auditory stimuli and overtraining at a critical period that is due to an executive deficit. Consistent with this interpretation, Heaton *et al.*'s (1998) finding of a greater memory for pitch in savant persons with autism might be due to the same factor and not, as interpreted by these authors, to a local bias in auditory information processing. However, the same explanation does not apply to Mottron *et al.*'s (in preparation) finding of superior performance in pitch modification by high-functioning persons with autism, as non-savant persons are not overtrained. As such, oversensitivity to auditory stimuli may exist primarily in persons with autism, independent of a local bias in the processing of information.

Concluding remarks

The majority of the recent literature regarding savant syndrome, including our own, assumed that special abilities could be adequately investigated with models that were built to account for the general clinical picture of autism. According to these existing cognitive models, the same cognitive abnormality responsible for the clinical picture of autism may also account for the development of a special ability (Mottron and Belleville, 1993; Frith and Happé, 1994). Conversely, it was presumed that close investigation of atypical information processing within a special ability could teach us about general information processing in autism. Current findings question the relationship between cognitive deficits and the emergence of a special ability, suggesting some modifications to this general schemata. This case highlights the importance of considering potential 'hidden variables' that may facilitate the appearance of a special ability. It also suggests that the developmental course of cognitive systems must be considered so that investigation of savant syndrome may facilitate our understanding of autism. Specifically, a critical period, as well as certain circumstances and predispositions, may be required for a cognitive deficit to cause the development of a special ability.

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Absolute pitch in autism: a case study

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Abstract

We present the case study of QC, a low-functioning adolescent with autism, who possesses the special ability of absolute pitch. QC participated in an indepth assessment of pitch perception and processing systems relevant for current cognitive models of autism. Her performance was compared to that of mental age- and chronological age-matched groups of persons with average intelligence, or to that of musicians. Absolute pitch in identification and production was confirmed. No abnormalities were found in perception of hierarchical (local-global) properties for visual patterns and music. However, a deficit in two components of executive functions, cognitive flexibility and planning for different materials, was evident. Short- and long-term memory for verbal, non-verbal and musical material were without particularities, although QC revealed an exceptional long-term memory for musical pieces when the piano was used for recall. This case suggests that absolute pitch in autism may not result from a multimodal deficit in processing global information. Rather, it may result from a lack of cognitive flexibility in a person with a marked interest for auditory stimuli that occurred at the critical age for the appearance of absolute pitch.

Journal

Neurocase 1999; 5: 485-501

Neurocase Reference Number: 0172

Primary diagnosis of interest

Autistic disorders

Author's designation of case

Key theoretical issue

- Does absolute pitch reveal a multimodal hierarchization information deficit when found in autism?
- Can special abilities be adequately investigated with models that were built to account for the general clinical picture of autism?

Key words: autism; special abilities; central coherence; hierarchization deficit; absolute pitch; music perception; executive functions

Scan, EEG and related measures

MRI

Standardized assessment

ADI, ADOS-G, WAIS, Raven, PPVT, Trail A & B, Wisconsin Card Sorting Test, Luria's Seriation, Spatial Reversal Task, Use of Object Test, Alternative Uses Test, Maze Task, Stroop, Continuous Performance Task, Boston Cancellation Task, Verbal Fluency, Buschke, Batterie informatisée d'évaluation de la mémoire Côte-des Neiges, Rey's 15 Words, Rey Figure, Birmingham Object Recognition Battery

Other assessment

Hierarchical perception for musical and visual material, short stories, digit and syllable span, phonological similarity effect, word length effect, alpha span, divided attention task, face span, spatial location span, musical recall and recognition

Lesion location

• None

Lesion type None

Language English