Music and Language Skills of Children with Williams Syndrome*

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ABSTRACT

We examined music and language abilities in a group of children with Williams syndrome (WS, $n = 19$) and a comparison group of normal children ($n = 19$) equivalent for receptive vocabulary. Consistent with previous reports and the model of Nonverbal Learning Disabilities (Rourke, 1989), the children with WS scored better on verbal than performance measures of the WISC-III, and performance on simpler verbal tasks (e.g., receptive vocabulary) was superior to performance on more complex verbal tasks (e.g., comprehension). Performance on music tests was *relatively* good, being comparable to mental age based on receptive vocabulary and similar to that of the comparison group. Music and language abilities were moderately correlated for both groups of children. Compared to normal children, the WS group expressed greater liking of music and a greater range of emotional responses to music.

The unusual constellation of characteristics that typifies individuals with Williams syndrome (WS) has captured the attention of scientists (e.g., Bellugi, Bihrle, Neville, Jernigan, & Doherty, 1992; Levitin & Bellugi, 1998; Mervis, Morris, Bertrand, & Robinson, 1999) and the popular media (e.g., the television program *60 Minutes*). Children with WS have 'elfin' facial features and are often described as friendly and talkative (Lowe, Henderson, Park, & McGreal, 1954; Mervis et al., 1999; Udwin, Yule, & Martin, 1987). Although these children are mentally retarded, they have an unusual cognitive profile with relatively preserved verbal abilities that contrast markedly with their extremely poor visuospatial skills (Bellugi et al., 1992; Lowe et al., 1954; Mervis et al., 1999; Udwin & Yule, 1991). Clinical, experimental, and anecdotal reports suggest that these children may also be relatively musical (Anonymous, 1985; Lenhoff, 1996; Levine, 1992; Levitin & Bellugi, 1998; Udwin et al., 1987; von Arnim & Engel, 1964).

For example, early descriptive studies reported that children with WS have good singing skills (von Arnim and Engel, 1964) and can easily learn songs (Udwin et al. 1987). More recently, Lenhoff (1996) provided a qualitative examination of the music skills of individuals attending a 'Music and Arts' camp for children with WS. The attendees displayed heightened levels of interest and emotional responsivity toward music, facility learning complex rhythms, excellent memory for lyrics, ease in composing song lyrics, ability with harmony, and an unusual number had absolute (perfect) pitch. In another study conducted at the same camp, Levitin and Bellugi (1998) administered a rhythm production test to 8 children with WS

(mean age = 13.4 years, *SD* = 3.6 years) and compared their performance to a control group of children 5 to 7 years of age. Although the groups were equivalent on the rhythm measure, the individuals with WS were more likely to produce musically 'compatible' rhythms when they responded in error.

Unfortunately, the WS group was a select cohort (music camp attendees) and groups were matched using reported norms on Piagetian conservation tasks without verifying that the groups were actually equivalent on these tasks. Thus, one cannot draw firm conclusions about the rhythm abilities of children with WS on the basis of this study. Nonetheless, the presence of relatively intact verbal skills combined with the possibility that such skills are accompanied by relatively intact musical skills provided the impetus for our research questions: (1) Are children with WS more musical than one would expect based on their overall cognitive abilities? and (2) How do the music skills of children with WS compare to their relatively intact language skills?

WS is a rare genetic anomaly characterized by a submicroscopic deletion on chromosome 7, which contains the genes for elastin, LIMK, and other genes (Ashkenas, 1996; Ewart, Jin, Atkinson, Morris, & Keating, 1994; Morris, Thomas, & Greenberg, 1993; Tassabehji et al., 1996). Individuals with WS exhibit vascular problems, such as supravalvar aortic stenosis and hypertension, that are associated with the loss of the elastin gene. The LIMK gene has been implicated in neural-cell development and is believed to be related to the deficits in visuospatial and visuoconstruction skills associated with WS. The overwhelming majority of children with WS (86 to 96%) also exhibit *hyperacusis*, which is characterized by aversive reactions to sounds that do not cause such reactions in normal individuals (Arnold, Yule, & Martin, 1985; Klein, Armstrong, Greer, & Brown, 1990; Udwin et al., 1987).

Although the neuropsychological profile associated with WS suggests right-hemisphere dysfunction, brain-imaging studies show mild microcephaly without specific lateralized structural lesions or anomalies (Jernigan, Doherty,

Hesselink, & Bellugi, 1993). Neocerebellar volumes and limbic structures are *relatively* large, however, and comparable to normal controls (Jernigan et al., 1993; Jernigan & Bellugi, 1990).

Exaggerated left-sided asymmetry of the planum temporale has been reported in professional musicians, particularly those with absolute pitch (Schlaug, Janke, Hunag, & Steinmetz, 1995). Because absolute pitch and relatively good musical skills have also been identified as possible characteristics associated with WS, Bellugi, Hickock, Jones, and Jernigan (1996) examined the planum temporale of individuals with WS. For the majority of their sample, the leftward asymmetry fell between the two groups of professional musicians (i.e., those with or without absolute pitch). Replication and extension of this finding could provide an anatomical basis for the reported musicality of individuals with WS.

Music

Musical skills are *not* typically associated with specific brain structures. *Amusia*, or the loss of music perception or performance abilities due to brain damage, is often accompanied by aphasia (Marin, 1982). In a review of 314 historical cases, Henschen (1920, cited in Judd, Gardner, & Geschwind, 1983) found that left-hemisphere damage and aphasia were present in 97% of cases of amusia. Because the cases of amusia included a wide range of deficits (music performance, composition, reading, music perception), the association with aphasia is too general to implicate localization of musical functioning. Indeed, results of neuropsychological investigations of musical abilities are often contradictory (Hodges, 1999), a likely consequence of the complex nature of music.

Nevertheless, research based on listening tasks, infant development, and lesion analysis reveals clues to the neurobiology and neuroanatomy of music processing. For example, perception of melodic contours (changes in pitch direction) is reliably associated with right-hemisphere functioning for both brain-damaged and normal individuals (see McKinnon & Schellenberg, 1997). By contrast, rhythmic processing

has been associated with left-hemisphere functioning, although such findings are inconsistent across studies (Peretz, 1990; Peretz & Morais, 1989). Despite the apparent links between aphasia and amusia, dissociations have also been reported. In a review of lesion studies, Sergent (1993) hypothesized that widely distributed, locally specialized neural substrates subserving music are proximate to, but distinct from, verbal areas. It is also possible that specific aspects of music and language share a common processor, with other aspects being independent. Indeed, a study of patients with amusia without aphasia demonstrated that processing of linguistic prosody and music were linked (Patel & Peretz, 1997).

In normal development, prosodic and melodic elements of speech and song directed to infants and young children are similar (Trehub & Trainor, 1998). Speech to infants typically involves exaggerated prosody with relatively slow and regular rhythmic patterns, shorter phrases, and greater repetition than adult-directed speech (Papousek & Papousek, 1981). It is also higher in pitch with simple pitch contours that span a greater range. Infant-directed singing is similarly slow, high-pitched, and rhythmically exaggerated (Trainor, Clark, Huntley, & Adams, 1997). Moreover, young infants prefer infant- over adult-directed speech (Cooper & Aslin, 1990; Fernald, 1985; Werker & McLeod, 1989), just as they prefer infant- over adult-directed singing (Trainor, 1996). Thus, adults' style of communicating in the auditory domain with very young listeners appears to capitalize on innate perceptual biases and preferences for particular auditory patterns. These similarities across domains are consistent with suggestions of a link between language and musical skills.

An explanatory framework for the unusual cognitive profile associated with WS is provided by Rourke (1989, 1995), who describes the syndrome of *Nonverbal Learning Disabilities* (NLD). NLD is thought to arise as the developmental outcome of an interaction between primary *assets* in auditory perception, rote learning, and simple motor skills, and primary *deficits* in tactile and visual perception, complex psychomotor skills, and adaptation to novelty.

During early development, a child typically explores the world through touching, feeling, seeing, and hearing. For a child with NLD, however, weak tactile and visual perception accompanied by difficulty with complex psychomotor skills render the world too confusing to assimilate through nonverbal processes. Instead, the child with NLD explores the world primarily with an auditory and verbally based approach. This unbalanced development results in an individual who has relative strength in skills subserved primarily by systems within the left cerebral hemisphere (e.g., simple language skills and auditory memory). By contrast, deficits are observed in skills that are thought to be subserved primarily by systems within the right hemisphere as well as in skills that require intermodal processing (e.g., nonverbal, fluid, or creative reasoning, abstract thinking, complex language comprehension, and prosody).

Although the overall level of abilities is lower for children with WS than for the typical child diagnosed with NLD, the pattern of assets and deficits is similar. Children with WS display relative strengths in auditory perception, verbal memory, speech articulation, and quantity of speech, but deficits in psychomotor coordination, visual-spatial-organization, and adaptation to novelty (Mervis et al., 1999). If one assumes that some aspects of auditory processing for language and music are shared, the NLD model also provides a framework for explaining relative strength in music as well as language. In other words, children with WS should demonstrate relatively good memory for simple and repetitive musical patterns in addition to their relatively good language abilities. Moreover, performance on simple language and music measures should be superior to performance on more complex measures. Some areas of reported strength for children with WS, however, such as facial memory and the perception and production of speech prosody (Udwin & Yule, 1991; Bellugi et al., 1988; Bellugi et al., 1994), are not consistent with the NLD profile and suggestive of relatively intact nonverbal functioning for this population in specific areas. Regardless, because these children appear to be 'tuned into' the melodies of speech (i.e., prosody), it is rea-

sonable to expect that their music-perception abilities might also be relatively good.

The linguistic abilities of children with WS are well established, yet their musical skills have not been empirically validated. On the one hand, some aspects of music abilities may be spared in this population relative to their overall level of cognitive functioning, as are some aspects of language use. Indeed, music skills in WS might even stand out as a strength in comparison to the normal population; if demonstrated, this would imply that WS comprises a group of musical *savants*. On the other hand, music may appear to be a relative strength simply because these children often respond enthusiastically to music.

The purpose of the present study was to examine the language and music abilities of children with WS. Because little is known specifically about music skills in individuals with WS, we focused on quantitative measurement of such skills. We expected that music and language skills would be correlated and that both would be better than nonverbal abilities. In addition, children with WS were expected to be similar to normal children of equivalent verbal level in terms of their musical abilities. Finally, if the cognitive profile associated with WS arises in the manner proposed for NLD (Rourke, 1989), intact auditory processing should be a primary means by which children with WS develop an understanding of the world. Thus, *interest* in music might also be greater for children with WS than for normal children.

To test the hypothesis that music and language skills represent areas of relative strength in children with WS, a comparison group with equivalent language skills was chosen. The choice of a language measure on which to equate groups was particularly important because children with WS are relatively strong in *simple* language skills yet weak in more complex areas such as language comprehension and pragmatics (Anderson & Rourke, 1995; Mervis et al., 1999; Rourke & Tsatsanis, 1996). For example, Verbal IQ as defined by the *Wechsler Intelligence Scale for Children, Third Edition* (*WISC-III*; Wechsler, 1991) measures simple *and* complex language skills and is unlikely to

provide an appropriate basis for comparison. By contrast, receptive vocabulary – a relatively simple verbal skill – better reflects the language strengths observed in children with WS. Thus, a measure of receptive vocabulary was chosen as the basis for establishing between-group equivalence.

METHOD

Participants
Children for the WS group were recruited from Williams Syndrome Associations in Canada and the United States through organization newsletters or meetings. Children for the comparison group were recruited through fliers posted at the University of Windsor and at local churches. All participants spoke English as their primary language and were without significant sensory or physical handicaps.

The study group consisted of 19 children with WS between 8 and 13 years of age (10 boys and 9 girls). Mean chronological age was 10 years, 6 months ($SD = 1$ year, 10 months) and mean mental age – calculated using scores on the *Peabody Picture Vocabulary Test-Revised* (*PPVT-R*; Dunn & Dunn, 1981) – was 8 years, 1 month ($SD = 2$ years, 2 months). The comparison group – equivalent for mental age on the PPVT-R – was selected from a larger group of 32 normal children between 5 and 12 years of age. Specifically, only children with PPVT-R standard scores between 85 and 115 (average range) were included. The resulting group of 19 children (11 boys and 8 girls) had a mean chronological age of 7 years, 11 months ($SD = 2$ years, 4 months) and mean mental age of 8 years, 1 month ($SD = 2$ years, 5 months), which was identical to that of the WS group.

Measures
Language and music skills were assessed through standardized measures, a questionnaire, and a semi-structured interview. Four language measures (in addition to the PPVT-R) were selected to evaluate a variety of skills – ranging from relatively simple to complex – in both groups of children. The *Auditory Closure Test* (Kass, 1964), arguably the least complex task, was included as a simple measure of sound blending. On this task, the child hears segmented sounds of a word and is asked to blend the sounds to produce the corresponding word. Verbal fluency, another relatively simple verbal task, was assessed through the *Controlled*

Oral Word Association to category (Animals) (Halperin, Healy, Zwitchik, Ludman, & Weinstein, 1989, as cited in Spreen & Strauss, 1991), which also provides a measure of *perseverations*. More complex aspects of verbal skill requiring auditory attention and working memory were measured with the *Digit Span* subtest from the WISC-III and *Sentence Repetition* (Spreen & Strauss, 1991). For the WS group only, overall verbal and nonverbal intellectual functioning was measured with the complete WISC-III. The *Comprehension* subtest of the WISC-III provided the most complex measure of language skills for the children with WS.

Our choice of measures to assess musical skills was limited because most music tests are designed to examine the skills of individuals undergoing formal music training. For our purposes, only skills present in infancy or acquired implicitly through exposure to music could provide a basis for the evaluation of general auditory pattern processing abilities. *Gordon's Primary Measures of Music Audiation* (*PMMA*) (Gordon, 1980) for children in kindergarten through third grade met our criteria. The test was standardized in 1978 on a sample of 873 children residing in suburban New York state. Test-retest reliabilities ranged from .73 to .76 with split-half reliabilities ranging from .72 to .86. The test has also been used to assess the music abilities of mentally retarded adults, with test-retest reliabilities at .81 or higher (Hoskins, Kvet, & Oubre, 1988).

The PMMA comprises *Tonal* and *Rhythm* subtests for which centile scores are computed. Each subtest has 40 taped trials. Each trial (both subtests) consists of a pair of short, monophonic melodic or rhythmic phrases, and children are asked to indicate whether the paired sequences are the same or different. *Standard* and *comparison* phrases are identical on *same* trials but not on *different* trials. *Tonal* phrases have 2 to 5 pure tones (sine waves) that differ in pitch but not in duration on 'different' trials; phrases are presented in either major or minor tonality. *Rhythm* phrases have 2 to 11 pure tones that vary in duration but not in pitch on 'different' trials; 'macro' or 'tempo' tones are included to help establish the meter but these are presented at a relatively low dynamic level and with a different timbre. Practice items are provided with each subtest.

A *Child Music Interest Interview* and a *Parent Music Questionnaire* were constructed for this study. Both contained questions pertaining to the children's musical interests, activities, knowledge, and environment. The parent questionnaire included additional items asking about the child's history of otitis media and hyperacusis.

Procedure

Participants were interviewed and tested at their convenience, either at their homes or at the University of Windsor. After children were interviewed about their musical background and interests, the two music and five language tests were administered in standardized order (i.e., Tonal, Sentence Repetition, Auditory Closure, Controlled Oral Word Association, Digit Span, Rhythm, and PPVT-R). When necessary, test order was modified to maintain the child's interest, but the Tonal subtest always preceded the Rhythm subtest. Parents completed a questionnaire concerning their child's musical and auditory history. For all but two of the WS group, a second session was required for administration of the WISC-III. For one child with WS, results from a recent psychological assessment (1 week prior to testing) were used; another child declined WISC-III testing. All children received a gift of a small toy upon completion of each session.

Administration of the Tonal and Rhythm subtests of the PMMA was adapted slightly to ensure understanding and to maintain interest. The subtests were introduced as 'Mr. Gordon's tests' and the child was asked, 'When is Mr. Gordon going to start?' before the tests began. Subtests were administered after five practice trials or perfect completion of one practice trial. For children who failed to understand the concepts of *same* and *different* (i.e., 4 children in the WS group, 2 children in the comparison group), *right* and *wrong* were substituted. Because of attentional difficulties, children in the WS group received more breaks and cues to attend than did children in the comparison group.

The Rhythm subtest proved to be especially difficult for a few children. Indeed, for 3 children with WS and 1 comparison child, the measure was unscoreable because of numerous 'don't know' responses or multiple responses per item. Accordingly, these children were given a score of 50% correct (chance level). No child scored below chance on the Tonal subtest.

RESULTS

Verbal Tests

The first set of analyses sought to confirm previously reported patterns of better verbal than visuospatial performance in children with WS. Table 1 provides means, standard deviations, and ranges for the WS group on WISC-III Fac-

tors, WISC-III IQ scores, and the PPVT-R. Consistent with previous findings, a paired t test revealed a superiority for verbal over visuospatial skills as measured by the WISC-III Verbal Comprehension and Perceptual Organization Factors, respectively, $t(17) = 7.42$, $p < .001$. The Verbal Comprehension and Perceptual Organization Factors of the WISC-III were used as additional measures of overall verbal and visuospatial functioning because they eliminate extraneous factors (e.g., mental arithmetic, graphomotor speed) present in the more commonly used VIQ and PIQ. Nonetheless, a comparison of VIQ and PIQ scores provided additional confirmation that the verbal skills of children with WS were superior to their visuospatial skills, $t(17) = 6.03$, $p < .001$. Indeed, the pattern of higher verbal than nonverbal performance was evident for all children with WS but one, for whom verbal and nonverbal performance were equal.

Additional analyses compared performance on the PPVT-R (a measure of relatively simple verbal skills) with performance on measures of more comprehensive and complex verbal skills, and with performance on nonverbal measures. Consistent with predictions, PPVT-R scores were significantly higher than Verbal Comprehension scores, $t(17) = 4.27$, $p < .001$, and VIQ scores, $t(17) = 5.53$, $p < .001$. Scores on the PPVT-R were also higher than Perceptual-Organization and PIQ scores, $t(17) = 6.43$, $p < .001$, and $t(17) = 7.35$, $p < .001$, respectively. Pairwise correlations between PPVT-R and WISC-III scores are provided in Table 2. Although correlations among Factor and related IQ scores and Full Scale IQ were expected, all correlations were significant, which implies that psychomet-

ric intelligence accounts for at least some of the variance across measures.

The next set of analyses tested further the hypothesis that the language abilities of the WS group would be inversely related to the complexity of the specific language task. Table 3 provides means and standard deviations for the nine verbal measures administered to the children with WS. A repeated-measures analysis of variance (ANOVA) on standardized scores confirmed that performance varied widely across measures, $F(8, 128) = 17.28$, $p < .001$, from within normal limits on the Auditory Closure task (the simplest task), to almost 3 standard deviations below the norm on the most complex language measure (WISC-III Comprehension).

By contrast, mean levels of performance for the comparison group (see Table 4) were within one standard deviation of the norm for each of the four verbal measures on which they were tested. A repeated-measures ANOVA with one within-subjects factor (standardized scores on the four verbal tests completed by both groups) and one between-subjects factor (WS vs. comparison group) revealed a significant interaction effect, $F(3,90) = 3.63$, $p = .016$, which confirmed that the WS and comparison groups exhibited different patterns of responding across the measures. Follow-up tests examined between-group differences separately for each of the four verbal measures. As shown in Table 4, the WS and comparison groups did not differ on the simplest tests (Auditory Closure and Controlled Oral Word Association). On measures requiring additional mental processing that involved attention and working memory (Sentence Memory and Digit Span), the comparison

Table 1. Scores of Children with Williams Syndrome.

	n	M	(SD)	Range
PPVT-R	19	77.53	(18.33)	48-109
Verbal Comprehension Factor*	18	65.44	(9.65)	50-84
Perceptual Organization Factor*	18	52.56	(3.94)	50-62
VIQ*	18	61.83	(10.27)	46-81
PIQ*	18	50.61	(4.84)	45-62
FSIQ*	18	52.72	(7.60)	40-69

Note. PPVT-R = Peabody Picture Vocabulary Test-Revised; * From the Wechsler Intelligence Scale for Children.

Table 2. Correlations between the Peabody Picture Vocabulary Test-Revised (PPVT-R) and the WISC-III Measures for Children with Williams Syndrome (*n* = 18).

	VC	PO	VIQ	PIQ	FSIQ
PPVT-R	.768	.490	.763	.628	.792
VC		.713	.982	.726	.969
PO			.649	.939	.783
VIQ				.669	.964
PIQ					.835

Note. VC = Verbal Comprehension factor; PO = Perceptual Organization factor. All correlations were significant, *p* < .05

group's performance was superior to that of the WS group, $t(36) = 2.42$, $p = .021$, and $t(36) = 3.51$, $p = .001$, respectively. Because of previously reported strengths in auditory memory (Anderson & Rourke, 1995; Mervis et al., 1999), performance on the Digit Span subtest was further analyzed to examine group differences on the less complex *forward* condition and the more complex *backward* condition. On both tasks, the comparison group's performance was significantly better than the WS group [forward: $t(36) = 3.14$, $p = .003$; backward: $t(36) = 3.10$, $p = .004$].

Music Tests
Means on the Tonal and Rhythm subtests of the PMMA are also provided in Table 4 for both groups of children. A repeated-measures ANOVA with one within-subjects factor (Tonal vs. Rhythm subtest) and one between subject-factor (WS vs. comparison group) revealed a main effect of subtest, $F(1,34) = 45.59$, $p < .001$. Both groups performed better on the Tonal subtest than on the Rhythm subtest [WS group: $t(17) = 6.19$, $p < .001$; comparison group: $t(17) = 3.45$, $p = .003$]. Consistent with predictions, the WS and comparison groups did not differ in their overall performance on the music tests. Although the interaction between group and subtest fell short of statistical significance, $F(1,34) = 2.92$, $p = .097$, between-group comparisons revealed similar scores on the Tonal subtest but a disadvantage for the WS group on the Rhythm subtest, $t(35) = 2.19$, $p = .036$. Performance on the Tonal and Rhythm subtests was highly correlated for the children with WS, $r =$

Table 3. Descriptive Statistics for Children with Williams Syndrome on the Verbal Measures.

	n	*M*	(*SD*)
Auditory Closure	19	–0.27	(1.13)
Controlled Oral Word Association	18	–0.78	(1.15)
PPVT-R	19	–1.50	(1.22)
Similarities*	18	–1.87	(0.88)
Digit Span*	19	–1.88	(0.75)
Vocabulary*	18	–1.98	(0.84)
Information*	18	–1.98	(0.85)
Sentence Memory	19	–2.25	(0.98)
Comprehension*	18	–2.74	(0.47)

Note. For comparison purposes, scores are converted to z-scores (number of standard deviations from the mean for normal children of the same mental age).
* WISC-III subtest.

.702, $N = 18$, $p = .001$, as it was for the children in the comparison group, $r = .773$, $N = 18$, $p < .001$.

To compare performance of the WS group on music measures with their performance on other measures, mental-age based z scores for the Tonal and Rhythm subtests of the PMMA were calculated for the 14 children whose mental age fell within the range of established norms (5.5 – 9.5 years). These children with WS had a mean mental age (based on PPVT-R performance) of 7 years, 5 months. Mean scores on the Tonal subtest (mental age z-score equivalent = .29) and the Rhythm subtest (mental age z-score equivalent = –.11) were not significantly different from 0, and, thus, typical for mental age. This finding provides additional confirmation that overall performance on the music subtests of the PMMA was consistent with simple verbal abilities, as predicted.

To compare performance of the WS group on music tests with their performance on other verbal and visuospatial measures, z scores for chronological age were *estimated* because test norms did not extend to the chronological age of the WS sample. To illustrate, consider a hypothetical child whose Tonal and Rhythm subtest scores were 0.5 standard deviations above the norm for mental age and 0.2 standard deviations below the norm, respectively, and whose raw PPVT-R score (from which mental age was derived) was 1.5 standard deviations below the norm for chronological age. For chronological age, then, this child's Tonal score was higher than her PPVT-R score but her Rhythm score was lower. To estimate her music abilities relative to chronological age, departures from the norm (i.e., z scores, or *SD* units) were summed. Thus, we estimated that this child would perform 1 (– 1.5 + .5) standard deviation below the norm for chronological age on the Tonal subtest, and –1.7 (– 1.5 + –.2) standard deviations below the norm for chronological age on the Rhythm subtest.

Mean estimated z scores for the Tonal and Rhythm subtests were –1.17 (*SD* = 1.19) and –1.75 (*SD* = 1.08), respectively, both of which are significantly lower than average (i.e., z score = 0), $t(13) = 3.69$, $p = .003$, and $t(13) = 6.06$, $p < .001$, respectively. The finding that tonal and rhythmic discrimination abilities – considered to be indicators of musical aptitude – are well below average for chronological age makes it clear that, as a group, children with WS are *not* musical savants. Rather, their music skills are *relatively* strong when compared to their marked deficits in other areas. Indeed, paired t tests confirmed that estimated z scores on the Tonal subtest were significantly higher than z scores on the WISC-III measures [Verbal Comprehension: $t(12) = 4.19$, $p = .001$; VIQ: $t(12) = 5.45$, $p < .001$; Perceptual Organization: $t(12) = 6.02$, $p < .001$; PIQ: $t(12) = –6.7$, $p < .001$; and FSIQ: $t(12) = 7.02$, $p < .001$]. Similarly, estimated z scores on the Rhythm subtest were significantly higher than Verbal Comprehension scores, $t(12) = 2.83$, $p = .015$, VIQ scores, $t(12) = 4.22$, $p = .001$, Perceptual Organization scores, $t(12) =$

Table 4. Scores on the Verbal and Music Measures.

	Williams		Comparison	
	M	(*SD*)	*M*	(*SD*)
Auditory Closure	12.90	(5.03)	10.68	(6.16)
Controlled Oral Word Association	11.79	(4.24)	13.90	(5.22)
Sentence Memory	10.48	(2.44)	12.84	(3.50)*
Digit Span	7.53	(2.50)	10.79	(3.19)*
Digit Span Forward	5.42	(1.35)	7.00	(1.73)*
Digit Span Backward	2.10	(1.37)	3.79	(1.93)*
Tonal	31.94	(5.80)	33.00	(6.17)
Rhythm	25.95	(4.38)	30.11	(6.99)*

Note. *Groups differed significantly, $p < .05$.

4.71, $p = .001$, PIQ scores, $t(12) = 5.37$, $p < .001$, and FSIQ scores, $t(12) = 5.83$, $p < .001$. (Thirteen of the children in the WS group had WISC-III results and z scores on the Tonal and Rhythm subtests.)

The next set of analyses evaluated the hypothesis that language and music skills would be correlated. Correlations between estimated z scores on the music tests and WISC-III scores for the WS group are provided in Table 5. Despite the relatively small sample, significant positive correlations were evident for the Tonal subtest (i.e., with FSIQ and VIQ) and the Rhythm subtest (i.e., with FSIQ, VIQ, and the Verbal Comprehension factor). In general, music abilities in our relatively small sample of children with WS were significantly associated with verbal abilities and overall intelligence but not with visuospatial abilities.

Correlations for the language measures completed by both groups are provided in Table 6. *Perseverations* on the Controlled Oral Word Association Test (considered pathological) are also included. Although the values in Table 6 provide suggestive evidence that the data from the WS group were somewhat more variable and less strongly correlated than those from the comparison group, statistical comparisons of the magnitude of the correlations revealed no differences between groups. For the groups combined, all measures of language were moderately correlated with the Tonal and Rhythm subtests of the PMMA. Subsequent analyses examined whether the observed associations between language and music skills may have been influenced by mediating variables. Specifically, a series of multiple regressions confirmed that associations between

language and music were still evident when differences in age, past history of otitis media, history of ear infection in the past year, past history of hearing loss, current or past history of music lessons, months of music study, or hours per week spent listening to music were held constant. In sum, moderately positive correlations between music and language skills were evident, as predicted, and such correlations were equivalent for both groups.

Musical Experience and Interest

Responses on the Child Music Interest Interviews and the Parent Language and Music Questionnaires were used to evaluate and compare groups in terms of their interest in music and their auditory characteristics. The groups did not differ in musical background or environment, or in their history of creating music. In both groups, almost all children (100% of the WS group and 84% of the comparison group) reported that music could make them feel happy. One child in the WS group responded negatively when asked if music could make him happy, but his response was scored affirmatively after he explained that music could not make him happy because it made him *more* than happy. Children in the WS and comparison groups differed, however, when asked if music could make them feel sad. Whereas 79% of the WS group responded affirmatively, only 47% of the comparison group made the same response, $\chi^2(1, N = 36) = 3.95$, $p = .047$.

Just as children with WS were moved to a greater range of emotions by music than children in the comparison group, they were also more extravagant in rating their overall feelings

Table 5. Correlations Between Standardized Scores on the Music and WISC-III Measures for Children with Williams Syndrome ($n = 13$).

	Tonal Subtest	Rhythm Subtest
Verbal Comprehension Factor	.465*	.599**
VIQ	.511**	.618**
Perceptual Organization Factor	.245	.193
PIQ	.364	.287
FSIQ	.552**	.603**

* $p = .055$, ** $p < .05$ (one-tailed tests)

Table 6. Correlations Between Raw Scores on Music and Language Measures for the Williams Syndrome Group, the Control Group, and the Groups Combined.

	Williams Syndrome Group		Control Group		Groups Combined	
	Tonal Subtest	Rhythm Subtest	Tonal Subtest	Rhythm Subtest	Tonal Subtest	Rhythm Subtest
PPVT-R	.374*	.395**	.736**	.507**	.579**	.436**
Auditory Closure	.375*	.426**	.684**	.490**	.520**	.373**
Digit Span	.386*	.261	.643**	.586**	.501**	.551**
Controlled Oral Word Association	.243	.167	.634**	.608**	.465**	.489**
Perseverations	−.740**	−.364*	−.358*	−.339*	−.606**	−.349**
Sentence Memory	.461**	.472**	.671**	.586**	.572**	.611**

$* p < .05$, $** p < .01$ (one-tailed tests).

for music. When asked to describe their interest in music on a scale from 1 (no interest) to 5 (love it), children in the WS group responded more enthusiastically ($M = 4.63$, $SD = .76$) than did those in the comparison group ($M = 3.95$, $SD = 1.08$), $t(36) = 2.26$, $p = .030$.

The data also corroborated previous reports of hyperacusis in children with WS. Indeed, a history of hyperacusis was evident for *all* of the WS group but for only 10% of the comparison group, $\chi^2(1, N = 38) = 30.76$, $p < .001$. Unusual fearfulness toward sound was also evident in *all* of the children with WS. Although this characteristic distinguished them from the comparison group, $\chi^2(1, N = 38) = 13.57$, $p < .001$, a substantial number of comparison children (47%) also had a history of unusual fear for certain sounds. Unusual liking for specific sounds also distinguished the groups, $\chi^2(1, N = 34) = 17.30$, $p < .001$; 75% of the children with WS but only one child in the comparison group exhibited an unusual liking for specific sounds. Because otitis media can affect auditory characteristics, the groups were compared on the basis of their history of otitis media, tubes in ears, and hearing loss. No differences between groups were found.

DISCUSSION

We examined the music and language skills of a group of children with WS and compared them to those of a group of normal children with equivalent levels of receptive vocabulary (as measured by the PPVT-R). The findings confirmed previous reports that children with WS perform better on verbal than on nonverbal tasks. In fact, performance on *all* nonverbal tasks, except for Picture Completion (a non-motor visuoperceptual task), was below performance on *all* verbal tasks. We also discovered that children with WS did particularly well (i.e., within the normal range) on tasks measuring relatively simple verbal abilities, but far less well (i.e., about 3 *SD*s below the mean) on measures of more complex language skills. In fact, performance of the children with WS on verbal measures was negatively associated with the complexity of the particular measure, a pattern that distinguished the two groups of children. These findings are completely consonant with predictions arising from the NLD model (Rourke, 1989).

Although it could be argued that the contrast between groups is simply a reflection of the difference in overall intellectual functioning (mentally retarded children are presumed to have difficulty with complexity), the pattern of performance for the WS group is also different from that of the majority of mentally retarded children, who typically perform better on nonverbal than on verbal tasks (Sattler, 1992).

A summary of the WS group's performance across all tasks (as assessed by z scores) is provided in Figure 1. Quantitative measures of music skills revealed that children with WS are relatively 'musical' – much like they are relatively 'linguistic' – with superior performance on the music tests compared to *all* nonverbal tasks and measures of complex verbal abilities. Within the limited range of music skills tested, the performance of children with WS was substantially better than one would predict based on their Full Scale, Verbal, or Performance IQs. Indeed, the music skills of children with WS were commensurate with their relatively strong receptive vocabulary (a relatively simple language skill). Moderate correlations between language and music skills were similar for both groups, which implies that simpler aspects of language and music skills are subserved by a common mecha-

nism that is used to process auditory patterns in the general population as well as in the WS population. The present results also suggest that for children with WS, *basic* pattern-perception skills in the auditory modality are stronger than their auditory rote-learning or working-memory abilities, despite the fact that auditory working memory tends to be a relative strength in the WS population (Mervis et al., 1999).

The findings of the present investigation are relevant to contemporary notions of *modularity* (Fodor, 1983; Jackendoff, 1987). According to Fodor (1983), the human mind consists of several *modules*, which are hard-wired areas specified for functioning in a particular domain. One obvious candidate for modularity is language (e.g., Chomsky's Language Acquisition Device). Whereas children with WS have relatively good language skills despite mild to moderate mental retardation in most cases, aphasics often exhibit good nonverbal skills despite their loss of language. As such, children with WS have often been viewed as the missing link in the search for a double dissociation between language and other abilities (e.g., Levitin & Bellugi, 1998). Although it is possible that *separate* music and language modules (see Jackendoff, 1987) are *both* relatively spared in
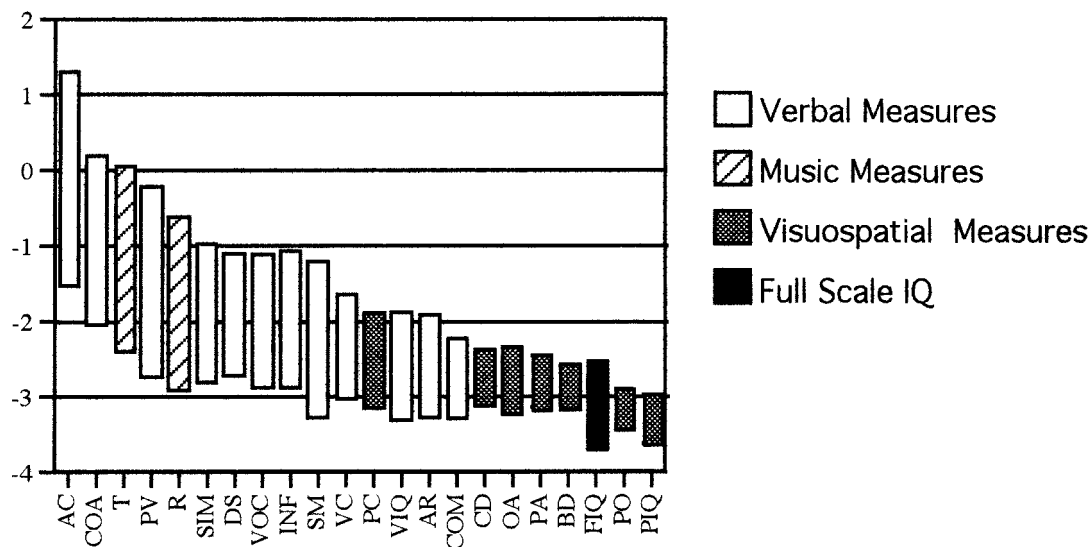

Fig. 1.   Mean performance (± 1 Standard Deviation) for the WS group on language, music, and visuospatial measures.

WS, we find it more parsimonious to speculate that basic abilities to process auditory patterns *in general* underlie the relatively good language *and* music abilities of children with WS. Indeed, because basic, general abilities are antithetical to the modularity concept, we interpret our results as providing rather strong evidence against the idea that WS is characterized by an intact language module or the idea that music and language are distinct modules.

Positive pairwise associations between verbal and nonverbal measures of the WISC-III and PPVT-R provided additional evidence against the idea of modularity of functioning in WS. Rather, the results indicated a role for psychometric intelligence, sometimes hypothesized to reflect general intelligence (*g*), such that children with better verbal and music skills tended to be less impaired overall, even on nonverbal measures. The somewhat greater variability on verbal measures compared to nonverbal measures could stem from floor effects on the nonverbal measures. Alternatively, there could be a greater range of ability in areas where children with WS often perform relatively well.

Even though absolute levels of performance were significantly lower, the WS group's pattern of performance largely paralleled that described by Rourke (1989) for individuals with NLD. Children in the WS group were functioning within the Moderately Mentally Retarded range for Full Scale IQ, Performance IQ, and the Perceptual Organization Factor (see Fig. 1). Scores on the VIQ and Verbal Comprehension Factor were slightly higher, in the Mildly Mentally Retarded Range. Performance on the less complex verbal measure of receptive vocabulary (PPVT-R) was even higher, with a mean score in the upper half of the Borderline range. Thus, our use of mental-age scores based on the PPVT-R – which gave relatively high estimates of verbal functioning – provided a conservative test of our hypothesis that music and verbal abilities would be equivalent.

Performance across language measures was related to task complexity, as predicted on the basis of the NLD model (Rourke, 1989). For the WS group, performance on the Auditory Closure test – arguably the least complex language measure – was closest to the norm. Basic auditory pattern perception is required for this task because it requires children to identify words that are presented in phonemic segments or clusters. Nonetheless, familiarity with the lexicon also affects performance. For example, when the segmented version of the word 'caterpillar' was presented, some children in both the WS and comparison groups identified the word before the last two sounds, 'l' and 'ar' were presented. Because 'caterpillar' is the only English word beginning with the sound pattern 'caterpi,' familiarity with the lexicon allowed these children to identify the word early.

Interestingly, some children in the WS group, but none in the comparison group, provided evidence of accurately synthesizing sound patterns despite lack of familiarity with the word. For example, one child, after correctly responding, 'tractor', asked, 'what is a tractor?' Another child, after responding correctly, asked, 'is that a word?' Children with WS may attend disproportionately to basic phonemic patterns compared to normal children, who may be more likely to rely on word familiarity. Recent reports of relative strength in phonological fluency among children with WS (Finegan et al., 1996; Mervis et al., 1996; Vicari et al., 1996; Volterra et al., 1996) lend support to the hypothesis that basic auditory pattern perception is intact in these children. Good phonological processing could also help to explain some parents' amazement at how fast their children with WS 'learn' to speak or sing in foreign languages. It is likely, however, that understanding of a foreign language would be very limited for these children.

According to the model of NLD (Rourke, 1989), secondary assets in auditory attention and memory are expected as an outcome of the over-reliance on primary assets in auditory perception and rote learning. For the WS group, performance levels on the Controlled Oral Word Association Test (Animal) were relatively good and similar to levels on the Auditory Closure Test (i.e., within 1 *SD* of the norm for chronological age). Because rote learning and basic auditory perception are considered primary assets within the NLD model, relatively good performance on this measure was expected.

Although semantic fluency on the word-association task was within the average range, performance of the WS group was inferior to that of the comparison group. This finding implies that semantic fluency and rote learning are less developed in children with WS compared to their basic auditory pattern perception skills. Indeed, poorer performance on measures that place even greater demands on auditory attention or working memory (i.e., Digit Span, Sentence Memory) provides further support for our suggestion that auditory memory is not as strong as simple auditory perception among children with WS. In individuals with WS, their overall level of cognitive functioning may limit the full development of the NLD syndrome such that skills dependent on primary assets in auditory perception are more fully developed than skills dependent on secondary and tertiary assets of auditory attention and rote memory. Nonetheless, the wide range of performance exhibited by the WS group on these measures leaves this speculation open to further investigation.

The present study is the first to provide compelling empirical evidence that music skills represent areas of relative strength for children with WS. Although music skills were at a level typical for mental age based on receptive vocabulary (also a relative strength), they were well below that expected for normal children of equivalent chronological age. In other words, musicality in children with WS may stand out in some instances to clinicians, parents, and teachers because of its *relative* strength, rather than because of high, absolute or savant-like levels of skill.

Although the performance of children with WS on the Tonal and Rhythm subtests was consistent with their mental age, scores on the Rhythm subtest were significantly lower than those for the comparison group. One explanation for this result concerns test order. In compliance with a standardized testing procedure, the Tonal subtest always preceded the Rhythm subtest. Unfortunately, children with WS exhibited notably increased attention difficulties on the Rhythm subtest that were not evident on the Tonal subtest. Similar problems were not observed in the comparison group. Thus, the decrease in Rhythm subtest scores for the WS group relative to the comparison group may have been caused by test order. This hypothesis could be examined in future research by counterbalancing the music subtests with testing order.

The relatively strong music skills of children with WS and the moderate correlations between music and language measures provide support for the hypothesis that relatively intact pattern-perception abilities in the auditory domain underlie *both* music and language skills in children with WS. For the WS group, this interpretation was further supported by the finding of relatively weak correlations between the music tests and measures of visuospatial or visuomotor abilities. It is also important to note, however, that the moderate correlations between language and music skills were equivalent for both groups, which implies that basic auditory-processing abilities underlie some aspects of music and language skills in the general population as well as in the WS population.

The overwhelming prevalence of hyperacusis among children with WS could mean that some aspects of their relatively normal music and language abilities are actually the product of *atypical* auditory processing. Unusual emotional responses to specific sounds were also characteristic of the children with WS and distinguished them from normal children. Specifically, all children in the WS group exhibited unusual fear of specific sounds, and almost two thirds exhibited unusual liking for specific sounds. Moreover, interest in music was higher in the WS group than in the comparison group, as we predicted. One possibility is that relatively intact auditory-perception abilities combined with large visuospatial deficits make auditory stimuli especially salient compared to other stimuli for children with WS. Such increased salience could make it relatively likely for children with WS to have unusual and strong emotional responses to specific sounds, or abnormal sensitivity to particular auditory stimuli. This perspective is similar to the developmental trajectory described by Rourke (1989) for children with NLD.

Questionnaire and interview responses provided additional evidence that emotional responsivity to music may be related to emo-

tional responsivity to sound in general. Although not questioned specifically, 37% of children in the WS group were described by their parents as having an ambivalent relationship with certain sounds. For example, despite extreme fear and anxiety regarding the school fire alarm reported for one child, this same child had an extreme fascination with the same sound when he was placed in charge of initiating the alarm for fire drills. Intense emotional reactions to music were also reported for some children in the WS group. For 2 of the children with WS, a specific love/hate relationship with music was described. For 7 children, an unusual love and fascination with music were reported. Unusual negative reactions to music were described for another 4 children. As babies, these children were reported to scream or cry uncontrollably when they heard lullabies or slow 'relaxing' music. Thus, the music skills of children with WS may develop along a pathway assisted by relatively intact auditory perception abilities, accompanied by fascination and heightened emotional sensitivity.

Implications for Musical Development in Children with WS

The enthusiasm and emotional responsivity to music demonstrated by children in the WS group combined with their relatively intact music abilities raises the possibility that purposeful development of musical skills could help to enrich the lives of these children. It should be noted, however, that there was considerable individual variability in responses toward music. Although most children in the WS group responded enthusiastically toward music, 2 children were notably indifferent toward music and 1 child expressed intense dislike of music. Thus, it would be unwise to apply suggestions for musical development indiscriminately to all children with WS.

In general, physical and cognitive limitations of children with WS are expected to impede skill development in many areas, including music performance. For example, difficulties with fine motor coordination would limit the choices of an appropriate musical instrument for such a child. Bowed stringed instruments generally require high levels of fine motor control to produce

even the simplest recognizable song and would be particularly poor choices for instrumental instruction. Guitars require fewer fine motor skills (fretting aids finger placement) and have been a successful choice of instrument for some children with WS when alternative (open) tunings and bar chords were used (National Williams Syndrome Association, 1997); additional instruments reported to be played successfully by children with WS include keyboards, pianos, drums, harmonicas, and trombones. Alternatively, because articulation skills are intact, singing might be an excellent route for the development of musical skill.

Because most children with WS are mentally retarded and because their cognitive profile parallels that of children with NLD (Rourke, 1989), we speculate that music instruction would be most useful if it involved simple tasks, imitation, and an abundance of repetition. Visuospatial-visuomotor impairments and difficulties with complexity are likely to make reading music an unrealistic goal. The fatigue and attentional difficulties noted during testing indicate that teaching sessions should be kept short with breaks provided as necessary. Minimizing distractions and establishing a predictable sequence of events during the music lesson could help to focus and maintain the child's attention.

Findings of creativity and emotional responsivity toward music in children with WS provide additional sources of potential on which musical instruction could be based. For example, challenging a child to create a song using a specific sound, feeling, or technique may help to motivate musical growth. In addition, focusing attention on the sound quality and the emotional aspects of music may help the child to develop productive *and* receptive musical-expression abilities. Because sound preferences, aversions, and emotional responses to music were quite personalized in our sample, these suggestions for teaching may need to be individualized to help each child gain the most from his or her musical experiences.

Limitations
Because our primary goal was to evaluate the level of music skills among children with WS, it

was necessary to use standardized measures of such skills. Only a few measures proved to be appropriate for assessing musically untrained children. Consequently, only very basic melodic and rhythmic discrimination skills were assessed. Two limitations involving the administration of the music tests were also observed. One concerned the standardized testing order for the subtests noted earlier. In addition, all children received additional (non-standardized) practice trials when necessary to ensure understanding of the task. Repeated practice trials may have influenced results by inducing practice or fatigue effects. Although between-group comparisons were not affected by this adaptation, comparison with normative data may be less valid than would otherwise be the case.

In a small clinical study such as this, sample size and subject selection are always limitations. Self-selection of subjects is a particularly serious limitation in the present context because the decision to participate in the study might have depended upon the child's interest in music. The possibility that this was an especially 'musical' subgroup from the WS population could not be assessed. Nonetheless, the WS and comparison groups were equivalent in musical background and environment. Moreover, because few children in either group had taken formal music lessons, 'formalized' musicality does not appear to have influenced interest in participating. Indeed, the possibility of self-selection based on musical interest applies equally to both groups and could not be eliminated without random sampling, which is virtually impossible in a study of this sort.

Conclusion

In conclusion, the present study provided an investigation of both a relatively well known and a new area of auditory processing in children with WS – language and music, respectively. The findings yielded evidence of relative strength in both simple music and language skills as predicted based on deductions from the NLD model. The overall pattern of results across verbal tests suggests that skills dependent on primary assets of auditory perception were superior to skills requiring secondary and ter-

tiary assets in auditory attention and memory. Overall, findings suggest that intact, basic auditory-processing abilities may underlie the music and language skills observed among these children. In addition, findings suggest that the NLD model may be extended to include relative proficiencies in some simple musical skills.

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