Is intonation impaired in children with Williams syndrome?

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1. Introduction

Williams syndrome (WS) is a rare genetic disorder with a prevalence of about 1 in 25,000 live births (Korenberg, Chen, Hirota, et al., 2000; Frangistakis, Ewart, Moris, et al., 1996). It occurs due to a microdeletion on chromosome 7, which results in a number of physical abnormalities, such as elevated blood calcium levels, sensitive hearing and high blood pressure, failure to thrive in infancy, abnormal sensitivity to certain classes of sounds (hypersacusis), and moderate to severe learning difficulties. WS has attracted considerable interest over the past couple of decades because of the unusual neuro-cognitive profile associated with individuals with WS. It has been argued that linguistic abilities are relatively strong, compared to general cognitive functioning and non-verbal abilities (Bellugi, Wang and Jernigan, 1994; Bellugi, Linchtenberger, Lai, and St.George, 2000; Clahsen and Almazan, 1998, 2001). However, recent research has begun to question the claim that linguistic abilities are strong in WS (Karmiloff-Smith, Grant, Berthoud et al., 1997; Grant, Valian and Karmiloff-Smith, 2002., Karmiloff-Smith, Brown, Grice et al., 2003; Stojanovik, Perkins and Howard, 2004). It seems that for many individuals with WS linguistic abilities are on a par with their general cognitive functioning. It has recently been shown that pragmatic abilities in the WS population may also be impaired (Laws and Bishop, 2004; Stojanovik, 2006).

Research has also shown that children with WS go through the language acquisition process in a different way to typically developing children. For example, Mervis and Bertrand (1997) reported that referential word production preceded referential pointing, and fast mapping of novel objects appeared prior to exhaustive sorting, which is the opposite of what is observed in typical development. Paterson, Brown, Gsodl, Johnson, and Karmiloff-Smith (1999) reported that infants and toddlers with WS have an atypical learning trajectory with regard to language and numeracy. In addition, Nazzi, Paterson and Karmiloff-Smith (2003) reported delayed phonological development in WS, which questions the strategies employed by individuals with WS to aid lexical development. Furthermore, Nazzi (2001) found that, contrary to the typical population, the ability to categorise is not correlated with lexical acquisition in WS.

Relatively little is known about intonation in this population. Anecdotal evidence suggests that individuals with WS have ‘odd’ intonation, however there has not been any systematic investigation of the expressive and receptive intonation abilities in this population. A study by Reilly, Klima and Bellugi (1990) reported abnormally high use of affective expressive prosody (pitch changes, vocalic lengthening and modifications in volume) by adolescents with WS. Receptive prosodic skills, however, were not investigated. A recent study by Catterall, Howard, Stojanovik, Szczerbinski and Wells (2006) tested two adolescents with WS using the manual version of Profiling Elements of Prosodic Systems for Children (PEPS-C) battery (Wells and Peppé, 2001). Both children had impaired expressive and receptive prosodic abilities and had pervasive difficulties, compared to chronological age (CA) matched controls, in all aspects of the understanding and production of prosody.

Using prosody and intonation effectively, and interpreting the prosodic features of people’s speech, is extremely important if communication is to be successful. Intonation is one of the suprasegmental aspects of speech, together with word and sentence stress, prosody and rhythm. It is commonly attributed four main functions (Roach, 2000): 1) the attitudinal function (affect), in which intonation is used to express emotional state or attitude; 2) the accentual function (focus), in which intonation is used to make specific words or syllables stand out in a stream of speech; 3) the grammatical function, in which intonation is used to discriminate grammatical units or, for example, disambiguate defining versus non-defining relative clauses, and indicate complex clause structure (chunking); and 4) the discourse function (interaction), in which intonation is used to regulate conversational behaviour, or to focus a listener’s attention on specific elements.

From a theoretical point of view, it has also been argued that prosody may underlie language abilities but currently, there is no consensus regarding the issue of whether intonation is independent of morpho-syntactic, segmental phonological and general cognitive impairments (Wells and Peppé, 2003). Weinert (1992) argued that German speaking children with Specific Language Impairment (SLI) did not use prosodic cues when learning rules in a miniature language, or in repeating sentences accurately, which suggested that prosodic deficits may be associated with language deficits. On the other hand, research by Snow (1998; 2001) showed that children with SLI were not poorer than age-matched controls on the use of final pitch movement and final lengthening to mark the end of a conversational turn, neither did they have any difficulty with imitating rising and falling intonation contours, suggesting that prosody was not associated with these children's language difficulties. A study by Wells and Peppé (2003) also showed that expressive and receptive language abilities and intonation performance in a group of children with speech and language impairments were not strongly related. Given that children with WS have been found to have difficulties with language structure which are comparable to those of children with SLI (Stojanovik, et al., 2004), the question to ask is whether language deficits in children with WS may be related to deficits in their intonation abilities.

The aims of the present study are: 1) to investigate the understanding and production of intonation of children with WS using specially designed software developed to assess comprehension and production of intonation in children (PEPS-C) by Peppe, McCann and Gibon (2003); 2) to investigate whether there is a correlation between receptive and expressive language abilities and non-verbal functioning and intonation skills; 3) to investigate intonation performance of children with WS in spontaneous speech and compare this to the intonation performance of TD children of a similar language and chronological age.

2. Method

2.1. Participants

A group of fifteen children with WS (7;06 to 13;09) were recruited through the WS Foundations in the UK. Their mean age was 9;06 and there were 4 males and 11 females. All the children had a positive FISH (fluorescent in situ hybridisation) test to confirm gene deletion and diagnosis of Williams syndrome. They also had English as their first language and no diagnosis of autism or hearing impairment. The performance of these individuals was compared to that of two control groups: a chronological age matched group (CA) and a receptive language abilities matched group (LA). The CA control group was composed of fifteen typically developing children aged between 8 and 12;04 with a mean age of 9;09. There were 4 males and 11 females. The LA group was composed of fifteen typically developing children with a mean age was 5;07 (range 4;03 – 7;04). The LA control group was matched to the WS group on raw scores of the Test for the Reception of Grammar (TROG 2) (Bishop, 2003). Each participant with WS was individually matched to one child from the LA group, all of equivalent or within one point raw score on the TROG. The difference between the mean raw score on the TROG for the LA and WS groups was non-significant ($U=79.5$, $p=.392$). A summary of the mean group's performance on the standardized language and non-verbal measures is presented in table 1.

Table 1: Group data on mean raw scores and standard deviations in the standardized measures

Group	[a]**TROG (raw)**	[b]**RCM (raw)**	[c]**ACE (SF) (raw)**
WS (n=15)	6.9 (sd.3.4)	14.4 (sd.4)	13.5 (sd.4.7)
CA (n=15)	19 (sd.0.9)	31.9 (sd.4.8)	31.2 (sd.1.2)
LA (n=15)	8.7 (sd.2.8)	19.5 (sd. 4.8)	23 (sd.6.6)

[a] TROG – Test for the Reception of Grammar (Bishop, 2003); [b]RCM – Coloured Progressive Matrices (Raven, 1982); [c]ACE (FS) – Syntactic Formulation subset of the Assessment of Comprehension and Expression (Adams et al., 2001).

2.2. Procedure

Each child was tested individually in a quiet room either at their school, home or at the university for two sessions. A session lasted approximately 45 minutes. All the children were administered the TROG as a measure of receptive language abilities, the Syntactic Formulation subtest of the Assessment of Comprehension and Expression (ACE) battery (Adams, Cooke, Crutchley, Hesketh, and Reeves, 2001) as a measure of expressive language ability and the Raven's Coloured Matrices (RCM) (Raven, 1982) as a measure of general non-verbal cognitive abilities. They were all asked to generate a story from a wordless picture book: *Frog, where are you* by Mercer Mayer. The children's narrative output was recorded.

Administration of the PEPS-C was preceded by a vocabulary check which was to ensure that the children were familiar with the vocabulary items which were to appear in the intonation tasks. Tasks were presented in random order to different participants to ensure that there were no presentation order effects. All the output tasks were recorded directly onto the laptop as well as on a DAT recorder as a backup.

2.2. Materials

2.2.1. PEPS-C battery

The updated computerised version of the PEPS-C battery was used in order to assess children's prosodic input (i.e. receptive) and output (i.e. expressive) skills (Peppé et al., 2003). The assessment follows a psycholinguistic framework (Wells and Peppé, 2001) and is based on an earlier manual version. The advantages of the updated computerised version of the PEPS-C software is that: 1) the computer records and stores judgments, responses and administrative details, and converts responses into scores which reduces the possibility of human error; 2) materials appear more quickly on the screen than the tester can produce cards which means that the administration time of the battery is reduced; 3) the computerised version allows for the randomisation of the order of stimuli and tasks which is not possible if stimuli are on audiocassette (Peppé and McCann, 2003).

The battery consists of 12 tasks (subtests). They are divided into form (bottom-up processing where no meaning is involved), which refers to auditory discrimination and voice skills required, and function (top-down processing which involves meaning), i.e. how communication is affected by prosody in speech. Each subtest consists of two practice items and 16 subtest items. The practice items ensure that the participant understands what is required in each task. A description of each subtest is included in Appendix 1.

The function tasks include: turn-end, affect, chunking and focus. These tasks involve identification in the input and production in the output tasks. The input tasks typically require the child to either select the picture which best corresponds to the way a word or utterance is being said (identification) whereas the output tasks (which are supported with pictures as well) require the child to use intonation in order to convey different meanings (production).

The form tasks include variations in intonation (short item) and prosodic variation (long item). These tasks involve same/different discrimination in input and imitation in the output tasks. In the input tasks, the child hears pairs of one or two syllable pitch patterns and is asked to say whether they were the same or different (discrimination). In the output tasks, the child is required to imitate the speaker.

2.2.2. Spontaneous speech

The recorded stories were analysed using Laryngograph hardware and associated software. The analysis was based on a 2-minute sample. The F0, mode, mean, minimum and maximum pitch and the pitch range were extracted. Laryngograph generates two types of measurements: DFx1 and DFx2. We used the DFx2 value, which is derived by including pitch points only when two successive vocal fold vibrations have the same frequency. To control for further error, such as that which might arise from pitch halving or doubling, we looked at the 90% range rather than the whole range for the minimum, maximum pitch and pitch range. Semitone conversion was used to normalize the pitch range data, as Nolan (2003) [x] suggests that a logarithmic scale best models speaker intuitions about pitch range. This allowed us to compare the pitch ranges of the subjects, even though they had different modal F0 and their pitch values were obtained from different ranges on the physical scale. The pitch range in semitones (ST) for each child was obtained using the following formula, in which f_{max} is the maximum pitch in Hz and f_{min} is the minimum pitch in Hz for a particular child:

$$(1) \quad st = 12\left(\frac{LN\left(f_{\max}/f_{\min}\right)}{LN2}\right)$$

3. Results

3.1. Results from the PEPS-C battery

Results from the PEPS-C tasks assessing intonation function are presented in Table 2. The scores are given in percentages correct, i.e. number correct out of number scorable items.

Table 2: PEPS-C Results – input tasks

	AI	**AO**	**CI**	**CO**	**FI**	**FO**	**TI**	**TO**
Group								
WS (n=15)								
Mean	72.9*	80.7*	61.8***	59.1***	54.9***	83.1***	86.1***	69.1**
SD	17.11	17.9	11.4	20.3	15.9	14.9	13.2	14.1
Minimum	50	56.25	43.7	28.6	37.5	57.1	56.2	53.3
Maximum	100	100	81.2	87.5	93.7	100	100	93.7
LA (n=15)								
Mean	85	92.3	75 [a]	68.6 [a]	66.2 [b]	93.7	78.7 [a]	62.8 [b]
SD	20	10.5	9.3	15.7	17.5	9.8	17.7	30
Minimum	43.7	68.7	62.5	43.7	37.5	68.7	43.7	18.7
Maximum	100	100	87.5	92.3	87.5	100	100	100
CA (n=15)								
Mean	87.5*	95.3*	89.2***/[a]	89*** /[a]	87***/[b]	97.5***	97.5***/[a]	87.9**/ [b]
SD	17.8	5	16.8	13.3	13	3.9	3.9	16.4
Minimum	43.7	81.2	50	60	62.5	87.5	87.5	56.2
Maximum	100	100	100	100	100	100	100	100

PEPS-C = Profiling Elements of Prosodic Systems – Child version; AI – Affect input; AO-Affect output; CI – Chunking input; CI-Chunking output; FI- Focus input; FO-Focus output; TI – Turn end input; TO – Turn end output. LA – controls matched on receptive language skills; CA – controls matched on chronological age

The differences between the WS and CA/LA groups are marked using stars * significance at 0.05; ** significance at 0.01 level; *** significance at 0.001 level; the differences between the CA and LA groups are marked using letters [a] – significance at 0.001; [b] – significance at 0.01; [c] – significance at 0.05

Due to heterogeneity of variance and non-normal distribution of the data, nonparametric tests were carried out. Kruskal-Wallis, the non-parametric equivalent to ANOVA, was used in order to examine differences between the groups and there was a main effect of group for Chunking (input and output), Focus (input and output) and Turn-end (input and output). Post hoc Mann-Whitney tests were carried out in order to investigate any significant differences between the three groups of participants. There were no significant differences between the WS group and the LA group on any of the function tasks. However, when compared to the CA matched children, the WS group was significantly different on all the function tasks. There were also significant differences between the CA and LA matched children on the Chunking input and output tasks, Focus input, Turn-end input and output tasks. In fact, the scores in table 3 also show that the LA children performed below chance on the chunking output, focus input and turn-end output tasks.

Results from the intonation tasks testing form are shown in Table 3. As above, scores are given in percentages so that they can be compared to those of the function tasks. Kruskal-Wallis was also used to examine differences between the groups in terms of their performance on the intonation tasks testing form.

Table 3. Raw scores in percentages (number correct/number scorable) on the FORM PEPS-C tasks

	SD	**SI**	**LD**	**LI**
Group				
WS (n=15)				
Mean	75***	82.8***	70.1***	69.4***
SD	21.4	10.8	23.1	20.2
Minimum	50	68.7	25	34.4
Maximum	100	96.9	100	90.6
LA (n=15)				
Mean	85.6 [b]	87.4 [a]	84.4 [c]	90.5***/ [b]
SD	11.8	13	10.3	6
Minimum	62.5	53.1	68.7	81.2
Maximum	100	96.9	100	96.9
CA (n=15)				
Mean	96.2***/ [b]	98.1***/[a]	93.7***/[c]	97.7***/ [b]
SD	6.2	3	5.8	4.2
Minimum	81.2	90.6	87.5	87.5
Maximum	100	100	100	100

SD – short-item discrimination; SI – short-item imitation; LD – long-item discrimination; LI - long-item imitation
* significance at 0.05; ** significance at 0.01 level; *** significance at 0.001 level. The differences between the WS and CA/LA groups are marked using stars: * significance at 0.05; ** significance at 0.01 level; *** significance at 0.001 level. The differences between the CA and LA groups are marked using letters [a] – significance at 0.001; [b] – significance at 0.01; [c] – significance at 0.05

The only significant difference between the WS group and the LA controls was on the Long-item imitation task ($p<0.001$). There were significant differences between the WS and the CA groups on all aspects of intonation form. There were also significant differences between the CA and LA groups on all four intonation form tasks.

3.2. Results from the correlations

In order to address the second research question, i.e. whether level of linguistic performance may be related to performance on the understanding and production of intonation, Spearman's correlations were carried out separately for the WS group and the control groups. The significant correlations for the two typically developing groups are presented in Table 4.

Table 4. Statistically significant correlations of prosodic tasks with [a]ACE-SF subtest and [b]TROG

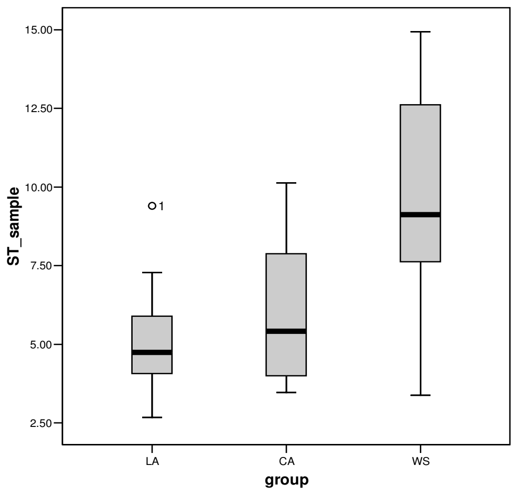
	Input			Output		
	C	F	T	C	SI	LI
[a]TROG	.402*	.517***	.538**	.450*	.451*	.433*
[b]ACE	.450*	.526**				

*p<0.05; ** p<0.01; ***p<0.001; C- chunking; F – focus; T-turn-end; SI-short-item imitation; LI – long-item imitation; [a]TROG – Test for the Reception of Grammar; [b]ACE-Assessment of Comprehension and Expression – Syntactic Formulation.

3.3. Results from spontaneous speech

Figure 1 shows the pitch ranges for each of the three groups. One-way ANOVA and Bonferroni post-hoc tests were carried out in order to investigate whether there were significant differences between the three groups. There was no significant difference between the LA and CA group. There was, however, a statistically significant difference between the participants with WS and their CA-matched peers of $p<0.01$, and also a significant difference between the LA-matched group and the WS group of $p<0.001$.

Figure 1: Pitch range data for the three groups of participants measured in semitones.



4. Discussion

The aims of the current study were: 1) to investigate the understanding and production of intonation of children with WS using specially designed software developed to assess comprehension and production of intonation in children (PEPS-C); 2) to investigate whether there is a correlation between receptive and expressive language abilities and non-verbal functioning and intonation skills; 3) to investigate intonation performance of children with WS in spontaneous speech and compare this to the intonation performance of TD children of a similar language and chronological age.

With regard to the first research questions, the results from the PEPS-C battery indicated that the children with WS did not differ from children matched for receptive language abilities on any of the expressive and receptive intonation tasks, apart from the Long-item imitation task. In this task, the participant is asked to imitate various prosodic patterns and the task assesses the ability to perceive and to imitate different prosodic utterances without a requirement for understanding of the forms. The difficulty that the children with WS had with this task, as indicated by the figures, might suggest difficulties with intonation production at a phonetic level rather than the linguistic/phonological level. It could be that the children with WS have specific difficulty with retaining pitch pattern information over longer prosodic domains in instances where no linguistic context is present. It has been shown that children with SLI have difficulties with processing over long prosodic domains which, it has been argued, may be associated with the short term memory deficits found in children with SLI (Wells and Peppé, 2003). Therefore, one possible explanation for the difficulties that the participants with WS had on this task could be that some underlying memory difficulties may be responsible for these children's difficulties with repeating prosodic variations. However, recent studies have shown that children with WS do not have short-term memory deficits, which is why a short-term memory test was not included in the methodology of the current study. Phonological short term memory, as measured with a test of non-word repetition, has been reported to be relatively good in WS (Grant et al., 1997; Majerus, Barisnikov, Vuillemin, Poncelet and van der Linden, 2003). Relatively good verbal short-term and working memory in WS has also been reported in a study by Robinson, Mervis and Robinson (2003). Given these reports it is unlikely that short-term memory could be the reason why the children with WS in the current study had difficulties with retaining pitch pattern information over longer prosodic domains.

Furthermore, although no measures of verbal working or short term memory were taken in the current study, closer inspection of the data shows that the majority of the children with WS (i.e. 12 out of the 14 participants) did not have difficulties with recalling the actual phrases, but they found it difficult to repeat them with exactly the same prosodic pattern. Therefore, memory deficits per se do not seem to be the reason for the children's failure in this task.

Another possible explanation for the difficulty that children with WS had with repeating various prosodic variations may be that these children had limited expressive language abilities. Close inspection of the data shows that the two participants who had most difficulty with the Long-item imitation task did not have lower expressive language scores than the remaining twelve participants and therefore it seems unlikely that their inability to repeat the various prosodic patterns was strongly associated with their expressive language skills.

Hence, the most plausible explanation for the difficulties that the children with WS had when having to reproduce various prosodic patterns as heard is due to a specific deficit in this prosodic domain. This suggests that some formal aspects of intonation may be dissociated from language and general cognitive abilities. Given that this is a task which only assesses form, it is possible that the lack of functional communicative value to the utterance contributes to the children's difficulty with repeating it as heard. As the results showed, on all of the function tasks, which assess the child's ability to understand and use intonation in communicatively meaningful ways, the children with WS performed similarly to the LA matched controls. Any pragmatic difficulties that have been reported for this population mean the way children with WS use or interpret intonation for meaning may be different from what would be expected for their chronological age and thus perceived as atypical.

On the other hand, the fact that children with WS are able to use intonation in communicatively functional ways, as well as interpret the communicative messages expressed through variations in intonation in line with their receptive language skills, could be one of the reasons why some studies have claimed that children with WS have relatively good or spared linguistic and communication abilities (Bellugi et al., 1994; Jones et al.,2000). This is by no means to say that imitating various prosodic patterns was the most obvious weakness in the intonation profile of children with WS. They had much lower average scores on the Chunking input and output tasks and on the Focus input task; however scores on these tasks were below chance for the LA controls as well, which means that these were not areas of particular difficulties for the WS group only, but also for the younger typically developing children. In fact, there were significant differences between the CA and LA children on all the tasks testing form and a number of intonation tasks testing various communicative functions conveyed by intonation, namely Chunking output, Focus input and Turn-end output. This corresponds to previous research which has shown that some aspects of comprehension and production of intonation develop throughout the school years. For example, with regard to the Chunking output task, Katz, Beach, Jenouri and Verma (1996), using a similar task to the one used in the PEPS-C battery, showed that children aged 5 and 7 years were unable to control duration and fundamental frequency in order to disambiguate syntactically ambiguous phrases. However Beach, Katz and Skowronski (1996) showed that children of the same chronological age were able to use prosodic phrasing in order to interpret grammatically ambiguous coordinated adjectival phrases of the kind used in the PEPS-C tasks as well [(pink and green) and white] and [pink and (green and white)].

With regard to focus, Cutler and Swinney (1987) have shown that children aged 3;0 to 5;11 can use accent placement to manipulate narrow focus in their own speech. Studies of comprehension, however, have shown that some aspects of interaction between accent and focus may not be mastered by the age of ten (Cruttenden, 1985). Therefore, these findings are well in line with the findings of our study which showed that the LA matched children (who were the ages of 4;03 and 7;04) performed below chance levels on the comprehension of focus.

There is no clear developmental picture about children's use of intonation for the purposes of interaction (Turn-end tasks) (Wells, Peppé and Goulandris, 2004). The children with WS in our study scored slightly higher than the LA controls, although the difference between the two groups was not statistically significant. In fact, there was a much larger variation amongst the LA children (minimum score was 18.7 and the maximum a 100) in comparison to the WS group. Other studies investigating this aspect of intonation have found a great deal of variation within typically developing children as well (Flax, Lahey, Harris and Boothroyd, 1991). The children with WS may have been more consistent with their responses than the LA children because they were older. Our current finding is consistent with the results obtained by Wells et al. (2004), which showed that children younger than 8 perform below chance on this aspect of intonation production.

With regard to the development of formal aspects of intonation as assessed on the Short and Long item discrimination and imitation tasks, the LA children, although scoring significantly lower than the CA children, were above chance on all the tasks, with relatively small standard deviations, showing a definite upward developmental trend. Although no data of this kind are available in the current literature, it seems to be the case that some aspects of intonation form related to auditory discrimination and voice skills are still developing throughout the school years.

The results and the subsequent analyses showed highly significant differences between the WS group and the

CA on all intonation tasks. Therefore, when compared to their chronological age peers, the children with WS may appear different in the way they use intonation as well as the way they interpret intonation as used by other people. This may be one of the reasons why children with WS have been perceived as having 'odd' sounding prosody. Having the CA group as a control group revealed a very important fact for therapeutic assessment and management, which is that understanding and production of intonation in children with WS is considerably below what would be expected of their chronological age and therefore a weakness in their linguistic profile relative to their chronological age. However, relative to their level of language comprehension, intonation abilities do not seem to be a particular weakness.

The second research question was whether intonation performance may be related to other language abilities. This is related to a more theoretical issue of the independence of intonation skills from other linguistic abilities. It should be pointed out that the intonation tasks in the PEPS-C battery have not been designed to focus on specific prosodic and rhythmic cues to language structure and therefore there is no direct correspondence between the expressive language tasks and the output tasks, and between the receptive language and the input tasks. Although it may look as if, for example, the Chunking task is designed to focus on specific prosodic cues to language structure (using stress to disambiguate single from compound nouns), no task on the ACE test focuses on this particular structure. As shown in previous studies (Local, 1980; Crystal, 1986; Wells, et al., 2004) aspects of intonation performance and language abilities in typically developing children are highly correlated. Wells et al., (2004), using an earlier version of the PEPS-C battery (Wells and Peppé, 2001), reported that expressive language skills as measured by the Formulated Sentences subtest of the Clinical Evaluation of Language Fundamentals battery (Semel, Wiig, and Secord, 1987) correlated highly with Chunking, Interaction[1] and Focus input task and Affect and Interaction output tasks. Performance on the TROG was highly correlated with all the input function tasks (Affect, Chunking, Interaction and Focus). The general conclusion of this study was that measures on the input tasks correlated strongly with measures of receptive and expressive language development, which means that during the school years, intonation and prosodic competence develops in line with other aspects of grammatical comprehension and production. Our findings for the two control groups in the present study are in line with previous findings in that performance on some functional and formal aspects of intonation was related to the development of other aspects of language.

However, this was not the case in the WS group, namely, almost none of the intonation tasks correlated with the WS children's performance on the language tests, apart from the Affect input task which correlated with a measure of receptive language ability (TROG). The lack of correlations between language and intonation abilities suggests that in individuals with WS linguistic and intonation abilities may not support each other in the same way as they do in the typically developing children, although participants with WS seem to be able to comprehend and produce intonation for communicative function as one would expect given the level of their receptive language skills as measured on the TROG. This finding is in agreement with the neuro-developmental model (Karmiloff-Smith, 1998) according to which WS shows an atypical developmental trajectory. An atypical developmental trajectory in WS for various cognitive skills has been reported in a number of different studies, as already mentioned in the introduction. Therefore, it could be the case that, although intonation skills in WS seem to correspond to the level of development of their language comprehension skills, the two aspects of linguistic abilities are not related to each other, which is probably due to altered constraints on cognitive and linguistic development (Karmiloff-Smith, 1998).

The spontaneous speech analyses using a laryngograph showed that the children with WS had a significantly wider pitch range than their chronologically and language matched peers. Research has shown that fundamental frequency and pitch are related to expressing emotions in speech (see Murray and Arnott, 1993 for a detailed review). This is in line with some previous findings that children with WS sound more emotionally involved (Reilly et al., 1990). The extremely wide pitch range found in the WS group may be one of the explanations why these children's prosody is often anecdotally described as 'odd'. Although we've found this difference in the *production* when measuring pitch range with electronic equipment, it would be interesting to interesting to find out whether naïve listeners actually *perceive* a difference in the emotional involvement between the children with WS and their CA and LA peers. Although on the Affect tasks of the PEPS-C battery the children with WS performed similarly to the LA controls, in fact, in spontaneous speech, they showed a profile which suggests something atypical about their pitch, which needs further investigation.

In conclusion, the intonation of children with WS does not seem to be impaired relative to their receptive language skills, although it is impaired relative to their chronological age. It also seems to be the case that, because there were almost no correlations between language and intonation abilities in the WS participants in the present study, intonation and other linguistic abilities in WS are not be strongly related. This means that children with WS do

[1] "Interaction" is referred to as "Turn-end" in the current version of PEPS-C.

not make use of intonation cues for interpreting or using language in the same way, or to the same extent, compared to typically developing children. In fact, research has shown that children with WS have delayed phonological speech perception skills (Nazzi, et al., 2003), which suggests that the developmental trajectory for speech perception in WS may be atypical and further research is needed in order to determine what this trajectory may be and how it may be related to the development of intonation skills. Unlike in the PEPS-C battery where the children with WS performed very similarly to younger typically developing children, the analysis of spontaneous speech showed a different picture. The children with WS were different from the younger typically developing children with regard to their use of pitch range, which may be one of the reasons why these children are perceived as having an 'odd' sounding intonation.

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APPENDIX 1- Description of the PEPS-C tasks

Function tasks

These tasks assess the child's ability to understand and produce intonation as used for communicative function

Turn end: Assesses the child's ability to understand and produce questioning versus declarative intonation. It assesses the function of intonation in interaction, by looking at single words constituted as conversational turns. The words are items of food and by the tone of the voice (a fall or a rise) the child is asked to decide whether an item was 'read', a fall, as opposed to 'offered', a rise. For the production task they had to produce the words by using appropriate intonation.

Affect: Assesses the child's ability to distinguish between 'liking' versus 'disliking' expressed in intonation on single words. Likes is conveyed by a rise-fall pitch contour whereas dislikes are conveyed by a fall-rise pitch contour. The stimuli involve opinions about foods and drinks. In the Input task the child was asked to express strong liking or disliking for food items that appeared on the screen. In the output task the child had to decide whether a voice on the computer liked or disliked a particular food item.

Chunking: Assesses the child's ability to perceive and produce grammatically ambiguous phrases which are disambiguated using prosody. The stimuli include pictures of food items and different-coloured socks. There are eight compound nouns and eight lists of two nouns. Chunking refers to prosodic delimitation of the utterance into units (or intonation phrases) for grammatical, semantic or pragmatic purposes. In the input task, the child hears a phrase, such as /FISH- FINGERS / AND FRUIT/ and is shown two pictures: one with two items on it (fish-fingers and fruit) and the other one with three items on it (fish, fingers and fruit). The child is asked to point to the picture that best corresponds with what the child has just heard. In this utterance the absence of a separate intonation phrase for FISH FINGERS makes 'fish-fingers' a compound noun. This means the utterance has two items and the child needs to point to the picture with two items only. Then the child may hear /FISH / FINGERS / AND FRUIT/. In this utterance, there are three intonation phrases, each with its own accent, and thus the utterance consists of a list of three food items and the child needs to point to the picture which has three items. Another example for the socks items is: /GREEN / AND PINK AND RED SOCKS/ vs. /GREEN AND PINK / AND RED SOCKS/. In the first utterance, the boundary after the first phrase means that the child should point to the picture which contains a pair of green socks and a pair of pink and red socks. If the child hears the second utterance, in which there is prosodic boundary after PINK, they need to point to the picture which contains a pair of green and pink socks and pair of red socks. In the output task, the child sees one picture only and is asked to say what they see. The output task thus assesses the ability to correctly produce a potentially ambiguous phrase using prosody.

Focus: Assesses the child's ability to identify and produce contrastive stress (tonicity). In the input task the children heard a sentence in which one of two colours was stressed. They were asked to say which one it was. For example: 'I wanted BLUE and white socks', BLUE was stressed and the child had to point to the blue picture. In the output task the children were presented with a football match between different coloured cows and sheep. Each time one of the animals had the ball the child would hear a voice (commentator) giving an erroneous description of the situation. E.g. for picture of a blue cow, the child would hear "the red cow has the ball". The child was asked to correct the commentator by saying: No, the BLUE cow has it! For each situation either the colour or the animal had to be changed depending on which of these the commentator got wrong, which assesses the child's ability to produce contrastive stress correctly.

Form tasks

These tasks assess the child's ability to perceive and produce various intonation and prosodic differences.

Short- item and Long-item discrimination tasks- These involve a same/different procedure. The child is presented with stimuli in a form where the lexical and grammatical information is not audible. Stimuli are laryngograph recordings, i.e. a recording of the laryngeal signal of the stimuli for input function tasks (Peppé and McCann, 2003, p.350). Pitch, loudness and length variations are preserved. The result is a 'buzz' - not dissimilar to listening to a speaker in an adjacent room, where the intonation is audible, but the content of the utterance is not.

Short-item and long-item imitation tasks- These tasks assess the child's ability to imitate different intonation or prosodic forms. For the short-item task the child heard a single word with a particular intonation pattern and was asked to repeat that word in exactly the same way as they heard it. For the long-item task the child heard an utterance with a particular prosodic form (e.g. GREEN and black socks) and was asked to repeat the phrase in exactly the same way as they heard it.

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