SPEECH PROSODY IN DOWN'S AND WILLIAMS SYNDROME: A COMPARISON

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ABSTRACT

Expressive and receptive speech prosody in a group of children with Williams syndrome and Down's syndrome are compared with each other and also with typically developing children using a computerized test battery. It is found that the WS children outperform the DS children on all expressive aspects of prosody, despite having comparable receptive language and non-verbal skills. This suggests that differences in prosodic ability may be linked to genetic disorder.

Keywords: prosody, Williams syndrome, Down's syndrome, atypical speech

1. INTRODUCTION

The study of populations affected by genetic disorders provides a unique opportunity to investigate the relationship between specific genotypes and different cognitive domains. These populations, in which the relationship between language and other cognitive functions might differ from what is seen in typical development, also provide a natural experiment to determine how different cognitive aspects may be related to each other. The present paper considers the relationship between prosodic abilities and receptive language and non-verbal skills in two distinct atypical populations: children with Williams syndrome (WS) and children with Down's syndrome (DS).

WS, which results from a microdeletion on one of the long arms of chromosome 7, is a relatively rare genetic disorder with a prevalence of 1 in 25,000 live births [7]. The deletion results in a number of physical abnormalities (e.g. elevated blood calcium levels, high blood pressure, failure to thrive in infancy, hyperacusis) and mild to moderate learning difficulties, with an IQ of between 40 and 60 on average [16]. Down syndrome (DS), which results from a genetic error in that the embryo receives three chromosomes 21, is a very common genetic disorder which occurs approximately in 1 in 800 live births [5]. It leads to a number of physical and cognitive abnormalities (e.g. short stature, hypothyroidism, hypotonia, congenital heart disease) and mild to moderate learning difficulties, with an average IQ range of between 40 and 60 [10]. Both groups present with uneven but qualitatively different profiles of cognitive abilities, despite similar levels of non-verbal IQ. DS individuals typically present with relatively good visuo-spatial abilities and poor expressive language skills [6]; speech and language are relatively more affected in individuals with DS compared with those with other learning difficulties [4]. In contrast, individuals with WS display relatively good expressive language abilities and poor visuo-spatial abilities [1]. Both disorders present with characteristic language profiles specific to each disorder. Thus in late childhood and adulthood, morpho-syntactic abilities, vocabulary skills and expressive prosodic skills are relative weaknesses in DS [9, 14]. In contrast, these abilities are on a par with general language and non-verbal abilities in WS [15].

2. PROSODY IN WILLIAMS AND DOWN'S SYNDROMES

The first published study of prosody in WS was a comparison with individuals with DS by Reilly, et al. [12], which reported that adolescents with WS used significantly more affective expressive prosody compared with the adolescents with DS who were matched on mental age to the WS individuals, and compared with two groups of typically developing (TD) children. The authors concluded that the use of affective expressive prosody by adolescents with WS was abnormal, as they used the same levels of expressive prosody regardless of how many times they told the same story, and irrespective of the audience – not the case with the individuals with DS and with the TD children. Little research on WS followed until the publication of a pilot study by Catterall, et al. [3]. This was the first study to investigate several aspects of both expressive and receptive prosodic

skills, and used specific experimental tasks from the manual version of the Profiling Elements of Prosody for Speech and Communication (PEPS-C) battery [17] (previously named Profiling Elements of Prodosic Systems – Children) to compare the prosodic skills of two adolescents with WS with those of two control groups, one matched for chronological age (CA), and one matched for language abilities (LA). The study reported impaired expressive and receptive prosodic abilities in both adolescents with WS when compared with the CA controls, but not with the LA controls. A larger-scale study [15], which included 14 children and teenagers with WS who were assessed on the computerised version of the PEPS-C battery [8], reported similar results. However, the children with WS had significant prosodic deficits in a number of prosodic domains when compared with TD CA controls, including the use of prosody to signal focus, to chunk phrases and to regulate conversational behaviour. The only task on which the WS group did not differ from the CA controls was a task which assessed the production and understanding of affect. However, a subsequent study involving the same participants [13] revealed that the WS group differed significantly from the TD controls in that their pitch range was much wider. This also contributed to the WS group being perceived as sounding much more emotionally involved when telling a story than the TD CA and LA controls.

Compared with research into prosody in WS, there is hardly any research in prosody in DS. The Reilly, et al. study mentioned above [12] reported that adolescents with DS made less use of affective expressive prosody in a story telling task compared with adolescents with WS and were more in line with mental age-matched TD children, but did not consider perception. Pettinato and Verhoeven [9] investigated the processing of word stress in children and adolescents with DS and reported disrupted stress structure in both production and perception. Stojanovik [15] reported that children with DS were particularly impaired on expressive aspects of prosody relative to language and non-verbal ability; receptive prosodic abilities were found to be matched to non-verbal abilities.

This paper addresses the following issues:

1. How do the receptive and expressive prosodic abilities of children with WS compare with those of children with DS?
2. Are there syndrome-specific prosody profiles linked to the two different genotypes?

3. METHODOLOGY

3.1. Participants

There were four groups of participants:

- Nine children with WS aged between 6 and 13;11 (mean 9;3) recruited through the Williams Syndrome Foundation in the UK.
- Nine children with DS aged between 8;2 and 12;11 (mean 9;3) recruited through DownsEd International in the UK.
- Eight younger typically developing (TD) children matched on non-verbal mental age (MA) to the WS and DS group aged between 4 and 7;7 (mean 5;3). The younger TD group was matched to the WS and DS groups on the basis of the scores from the Coloured Progressive Matrices (see below).
- Eight TD children matched on chronological age (CA) to the WS and DS groups and aged between 8;1 and 10;11 (mean 9;8).

The TD groups were recruited through local UK schools.

3.2. Materials

Prosody was assessed using the computerized version of the PEPS-C battery [8]. To assess language comprehension, all the participants were given a standardised language test, the Test for the Reception of Grammar (TROG) [2]; non-verbal cognitive abilities were assessed with a standardised non-verbal test, the Coloured Progressive Matrices (RCM) [11]. Table 1 shows the raw scores on the standardised tests for the groups.

Table 1: Participants' raw scores on standardised tests.

Group	*TROG raw*	*RCM (raw)*
DS (n=9)	5 (sd = 1)	15 (sd = 3)
WS (n=9)	8 (sd = 3)	15 (sd = 4)
TD MA (n=8)	6 (sd = 2)	16 (sd = 4)
TD CA (n=8)	19 (sd = 1)	33 (sd = 4)

The PEPS-C battery consists of six input (I) and output (O) tasks, four of which assess prosodic function – affect (AI; AO), chunking (CI; CO), focus (FI; FO), turn end (TI; TO) – and four of which assess prosodic form – short-item discrimination (SD) and imitation (SI) and long-

item discrimination (LD) and imitation (LI). For further description see [8].

3.3. Procedure

The TROG, RCM and PEPS-C battery were administered to individual participants either in a quiet room at their school, at homes or in a sound-treated room. The session lasted approximately 60-90 minutes with as many breaks as needed for the children to avoid fatigue. Each child was first administered a picture naming task to ensure that the children were familiar with the lexical items which appeared in the PEPS-C. Tasks were presented in a random order to different participants to control for presentation order effects. All the output tasks were recorded directly onto a laptop, and to DAT as a backup. Output tasks were recorded with the PEPS-C software by using a lapel microphone (Sony ECM 717) connected to a laptop. The tasks were recorded at a sampling frequency of 22.05 kHz.

The responses from 25 out of a total of 34 participants on the output tasks, initially rated by the researchers collecting the data, were independently rated by one of two trained phoneticians. Inter-rater agreement, calculated on a random 20% of this material using kappa coefficient, was very high (κ=.845; $p<0.001$).

4. RESULTS

The data were analysed using a One-Way ANOVA with posthoc Bonferroni comparisons if the variance was homogeneous (AI, CI and FI) or Tamhane's comparisons if the variance was non-homogeneous (all remaining variables). The results from the CO task are not presented here because seven out of nine participants in the DS group could not do the task. Significance is at $p\leq0.05$.

4.1. Function tasks

A main effect of group for all aspects of prosody function was revealed: AI ($p=0.046$); AO, CI, FI, FO, TI and TO ($p\leq0.000$). Post-hoc comparisons showed that the DS group was poorer than the CA group on the AI task, and the DS group was poorer than the WS, MA and CA groups on the AO task. Regarding the chunking tasks, the DS group was poorer than the CA and MA groups. On the FO task, both the WS and the DS groups were poorer than the CA controls, whereas on the FO task only the DS group was poorer than both the MA and CA controls. On the TI task, the DS group performed worse than all other three groups, while on the TO task the DS group was poorer compared to the WS and CA groups. For the results in tabular format with information on statistical significance, see image file 1.

4.2. Form tasks

The DS group was significantly poorer than all the other groups on the SD and SI and LD and LI tasks. See image file 2.

5. DISCUSSION

The prosodic profile of the DS children is clearly different from the prosodic profile of the WS children. The DS group showed overall poorer performance on all tasks, and in particular on the tasks assessing expressive prosody. With regard to prosody function, performance of the DS children was particularly low on the AO task, assessed by requiring the participant to express likes and dislikes of various types of food items. DS children were inconsistent with their production of the prosodic features associated with the expression of likes and dislikes in English (e.g. the rise-fall/fall-rise tone dichotomy modeled in the sample tasks), whereas the WS children were fairly consistent and better able to be identified by the raters as liking or disliking an item. Thus, despite the fact that both populations displayed a comparable level of receptive language and non-verbal abilities, the children with DS showed a significantly lower ability to produce affect intonation. This is in line with the findings of the very first study to compare expressive affective prosody in these two groups [12]. It seems the DS genotype particularly affects the ability to produce the prosodic information used in English to express this particular affective state; it is exclusively linked to the DS cognitive profile and does not seem to be related to non-verbal abilities and level of language understanding. If it had been, we would have expected the WS group to also have difficulties with this skill.

The other function which distinguished between the two groups was Turn-end. The WS participants outperformed the DS participants on both the TI and TO tasks. The DS participants had particular difficulty with TO: on average only 38% of items were correctly identified by the raters, suggesting that the DS children were unable to reliably signal the difference between questions and statements. The WS children, on the other

hand, were not only able to use the two different tones for this purpose, but they also, on average, performed better on this task than the MA TD children.

With regard to prosody form, the DS group performed significantly lower than the WS group and both TD groups. This applied to expressive and receptive prosody; when the task involves prosody perception or production without any communicative function, the individuals with DS tend to be weak and perform poorly, suggesting prosodic deficits in this population.

When compared with TD children, WS individuals show prosodic abilities which are similar to those of the MA-matched children and below those of the CA-matched children. The picture seems different for DS individuals, who seem to have a more varied and generally weaker prosodic profile in that some prosodic abilities seem to be in line with their mental age whereas others (expression of affect, understanding and producing questioning versus declarative intonation and prosody form) seem to be lower than expected for their mental age.

It should be noted that there was individual variation within the two atypical groups with 10% of the DS children and 20% of the WS children performing within norms, which is reflective of the heterogeneity often reported for disordered populations.

6. CONCLUSION

Children with WS and those with DS differ with regard to their prosodic abilities, despite having comparable language and non-verbal skills. Notably, WS children score higher than DS children on all expressive aspects of prosody under investigation and, in particular, on tasks assessing the affect and turn-end prosodic functions. The data point in the direction of the existence of two distinct prosodic profiles, each linked with a specific genetic disorder. In WS, prosodic skills are in line with other cognitive abilities, whereas in DS, the prosodic profile is mixed with severe weaknesses evident in some prosodic domains.

7. ACKNOWLEDGEMENTS

This research was partially supported by award RES-000-22-1302 from the UK ESRC.

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