

## Communication profiles of individuals with Down's syndrome, Angelman syndrome and pervasive developmental disorder

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### Abstract

The communication profiles of individuals with Down's syndrome (DS), Angelman syndrome (AS) and pervasive developmental disorder (PDD) were investigated and contrasted. Seventy-seven individuals participated in the study. A within-group analysis revealed that those with DS performed better on tacting or labelling and echoing than on manding or requesting. No other effects were found, apart from an absence of echoing in those with AS, a result that is hardly surprising. A between-groups analysis revealed no differences between the aetiological groups in terms of their use of mands or requests and tacts. Individuals with DS and PDD did not differ in their scores on echoic functioning. The implications of these findings for the study of behavioural phenotypes and for communication intervention are discussed.

**Keywords** Angelman syndrome, communication profiles, Down's syndrome, pervasive developmental disorder

### Introduction

Aetiology is increasingly supposed to account for differences in behaviour and development among individuals with intellectual disability (ID) (Dykens 1995). Differences have been found that can not be explained by variations in environment, education and training alone, although the indirect effects of genetic disorders on behaviour can not be discounted. The study of the relationship between aetiology and behaviour, i.e. behavioural phenotype, comprises the relationship between aetiology and adaptive behaviours (i.e. communication, academic and social behaviours), and between aetiology and behaviour disorders. Behavioural phenotype can best be regarded as 'the heightened probability that people with a given syndrome will exhibit certain behavioural and developmental sequelae' (Dykens 1995, p. 523).

In the early 1970s, a relationship between Lesch–Nyhan syndrome and self-injurious behaviour was suggested (Nyhan 1976), and this was later extended to relationships between Prader–Willi syndrome (PWS) and hyperphagia (Holland *et al.* 1995), PWS and psychotic symptoms (Clarke *et al.* 1998), Rett syndrome and stereotypic hand movements (Van Acker 1991), and Smith–Magenis syndrome and 'self-hugging' (Finucane *et al.* 1994), to

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mention but a few. Until now, these relationships have not been challenged, although a straightforward causal relationship between aetiology and behaviour is denied (Dykens 1995).

Individuals with Down's syndrome (DS) show a relative weakness in grammar, whereas pragmatics is an area of some strength (Chapman *et al.* 1998). Children with Angelman syndrome (AS) rarely acquire speech (e.g. Alvares & Downing 1998), while those with pervasive developmental disorder (PDD) have a high rate of immediate and delayed echolalia (e.g. Charlop 1986).

Within-aetiology studies have been most prevalent and have provided much useful information. However, comparisons across aetiological groups or studies between aetiologies have been conducted less frequently, but these are crucial to assess to what extent specific behavioural features are distinctive for a given aetiology. Fortunately, these studies are now emerging. With respect to behaviour disorders, Dykens & Kasari (1997) compared individuals with PWS, DS and non-specific intellectual disability, and Clarke & Boer (1998) compared individuals with PWS, Smith-Magenis syndrome and Cri-du-Chat syndrome.

With respect to cognitive aspects of behaviour, studies between aetiologies have revealed that individuals with DS have linguistic profiles that differ from individuals of an undifferentiated contrast group (Hodapp 1996). Individuals with DS also demonstrate relative strengths with respect to visual processing, while individuals with PWS and fragile-X syndrome share a weakness with regard to sequential processing (Scharfenaker *et al.* 1991). Children with Williams syndrome outrank those with DS in terms of linguistic development, especially at late age (Singer *et al.* 1997).

To gain more information in this area, various aetiological groups should be compared. An area that deserves special attention is aetiology as it is related to communicative functioning. To the extent that aetiology explains one's level and characteristics of communicative functioning, training and education are considered to have lesser impacts and are then likely to be withheld.

The present exploratory study addresses the question of whether aetiology-specific profiles exist in the area of communication. Since people with these aetiologies often participate in commu-

nicative behaviour training (i.e. speech and manual signs), the authors compared the communication profiles of individuals with DS, AS and PDD.

## Subjects and methods

The participants were 77 individuals with severe and moderate levels of ID, ages ranging from 3.2 to 52.2 years, who belonged to one of the three aetiological groups mentioned above. There were 26 participants with DS, 26 with AS and 25 with PDD. For the first two aetiological groups, diagnosis was based on cytogenetic procedures. Individuals with PDD were diagnosed (or a former diagnosis was reaffirmed) by a child psychiatrist, using criteria based on the *Diagnostic and Statistical Manual*, 4<sup>th</sup> edition (APA 1994). The participants were recruited through existing contacts between the University of Nijmegen, and a number of facilities, day-care centres and schools throughout the Netherlands. The participants lived at home (55%) or at a residential facility (45%).

The DS group had a mean chronological age (CA) of 11.7 years (range = 4.3–40.4 years) and a mean Adaptive Composite Behavior (ACB) score (Vineland Adaptive Behavior Scales; VABS) of 17 months (SD = 5.05 months). The AS group had a mean CA of 15.8 years (range = 3.2–52.2 years) and a mean ACB score of 16 months (SD = 4.4 months). The PDD group had a mean CA of 15.1 years (range = 5.5–38.6 years) and a mean ACB score of 22 months (SD = 6.13 months). However, because of logistical constraints, ACB scores were only obtained for a total of 43 participants from the three groups. Therefore, a relationship between ACB and Verbal Behavior Assessment Scale (VerBAS) scores was determined ( $r = 0.606$ ,  $P = 0.000$ ). This significant relationship allowed the present authors to extrapolate VABS scores to the 34 participants of the three groups for whom ACB scores were not available. This was conducted by using the missing values procedure (SPSS, Version 8.0). After conducting a one-way analysis of variance (ANOVA), statistically significant different ACB scores were found between the aetiological groups ( $F_{2,74} = 12.09$ ,  $P = 0.000$ ). Because of these differences mand, tact and echoic scores on the VerBAS

were adjusted by means of available and extrapolated VABS scores, using regression analysis to make the groups as comparable as possible. The resulting raw residual scores were used in the analysis. Parent and caregiver consent for the study were obtained.

The modality of the participants' communication differed. Participants with DS used speech (80%), and manual signs or communicative gestures (20%). None of them used other means of communication (e.g. pictures or pointing). None of the participants with AS used speech, 68% of them used manual signs and gestures, and 32% used other communication means. Participants with PDD used speech (78%), manual signs and gestures (22%), and none of them used other means of communication.

#### Recording

Data on communicative functioning were collected using the VerBAS (Duker 1999). This questionnaire is comprised of 15 items with scale values ranging from '0' to '4', based on frequency. The questionnaire estimates communicative functioning irrespective of modality, i.e. whether an individual uses either speech, signs, gestures or pointing. Inter-rater reliability and internal consistency have proven satisfactory (Duker 1999). Factor analysis has revealed three factors, accounting for 63.5% of the variance. Factors are labelled as manding, tacting and echoing, comprising three scales. Examples of items are: 'If s/he does not want the offered food, drink, or toy(s) any longer, does s/he say this, make the appropriate sign, point to the appropriate symbol, or pushes it away?' (i.e. mand); 'If you point to a body part that is known to her/him and ask: "What is this?" does s/he say, sign, or point the correct name of it?' (i.e. tact); and 'If you ask her/him a question, for example, "What do you want?", does s/he imitate the question exactly?' (i.e. echoic). The theoretical background of the VerBAS is Skinner's (1957) treatise on verbal behaviour. The questionnaire was individually filled out by the experimenter while interviewing the parent(s) or a caregiver (i.e. teacher or direct-care staff member) who had known the ratee for at least 6 months. Participants who endorsed a score of '0' on 13 or more items of the VerBAS were excluded from participation, as

well as those who endorsed a score of '4' on 13 or more items of this questionnaire.

## Results

### Within-group analysis

The raw residual scores obtained above were transformed into residual deviation scores for each participant. Then a series of univariate *F*-tests was conducted, revealing that participants with DS had a mean residual deviation score on the mand scale of  $-1.85$  ( $SD=3.48$ ) that significantly differed from zero ( $F_{1,74}=8.29$ ,  $P=0.005$ ). Their mean residual deviation score on the tact scale of  $1.26$  ( $SD=3.46$ ) also significantly differed from zero ( $F_{1,74}=4.35$ ,  $P=0.040$ ). Their mean residual deviation score on the echoic scale of  $0.59$  ( $SD=2.58$ ) failed to differ significantly from zero ( $F_{1,74}=1.33$ ,  $P=0.253$ ).

Participants with AS had a mean residual deviation score on the mand scale of  $1.88$  ( $SD=2.80$ ) that significantly differed from zero ( $F_{1,74}=8.52$ ,  $P=0.005$ ). Their mean residual deviation score on the tact scale of  $-0.33$  ( $SD=2.08$ ) failed to significantly differ from zero ( $F_{1,74}=0.29$ ,  $P=0.591$ ). The mean residual deviation score for this group on the echoic scale was  $-1.55$  ( $SD=2.49$ ), which significantly differed from zero ( $F_{1,74}=9.19$ ,  $P=0.003$ ).

Participants with PDD had a mean residual deviation score on the mand scale of  $-0.027$  ( $SD=3.53$ ), which failed to differ significantly from zero ( $F_{1,74}=0.00$ ,  $P=0.967$ ). Also, their mean residual deviation score of  $-0.97$  ( $SD=3.52$ ) on the tact scale failed to differ significantly from zero ( $F_{1,74}=2.49$ ,  $P=0.119$ ). Finally, the participants' mean residual deviation score on the echoic scale of  $1.00$  ( $SD=2.79$ ) approached significance ( $F_{1,74}=3.67$ ,  $P=0.059$ ).

Univariate *F*-tests were used to determine differences between mean residual scale values within each of the three aetiological groups.

As multiple comparisons were in effect, the Bonferroni correction was conducted by dividing the nominal alpha level by the number of comparisons with each single data set, resulting in a type I error level set at  $0.025$ . For participants with DS, the difference between these mean values for the

scales of mand and tact was statistically significant ( $F_{1,74}=7.48$ ,  $P=0.008$ ), with means of  $-1.02$  ( $SD=4.53$ ) and  $2.09$  ( $SD=5.79$ ), respectively. The difference between the mean residual scores on the mand and echoic scales was statistically significant ( $F_{1,74}=6.05$ ,  $P=0.016$ ), with means of  $-1.02$  ( $SD=4.53$ ) and  $1.42$  ( $SD=4.2$ ), respectively. The difference between the mean residual scores on the scales of tact and echoic failed to be statistically significant ( $F_{1,74}=0.54$ ,  $P=0.467$ ).

For participants with AS, the difference between residual mean values on the mand and tact scales failed to be statistically significant ( $F_{1,74}=3.75$ ,  $P=0.057$ ). The difference between mean residual scores on the mand and echoic scales was significant ( $F_{1,74}=11.94$ ,  $P=0.001$ ), with means of  $0.66$  ( $SD=3.94$ ) and  $-0.278$  ( $SD=2.3$ ), respectively. The difference between the residual mean values on the scales of tact and echoic failed to be statistically different ( $F_{1,74}=1.79$ ,  $P=0.186$ ).

For participants with PDD, mean residual values on the mand and tact scales failed to differ significantly ( $F_{1,74}=0.66$ ,  $P=0.418$ ). The difference between mean residual values on the mand and echoic scales also failed to differ significantly ( $F_{1,74}=1.03$ ,  $P=0.314$ ). Finally, these participants' values on the mean residual values of tact and echoic significantly failed to differ ( $F_{1,74}=4.45$ ,  $P=0.038$ ), with means of  $-0.57$  ( $SD=6.43$ ) and  $1.41$  ( $SD=4.71$ ), respectively.

#### Between-groups analysis

A  $3(\text{groups}) \times 3(\text{scales})$  overall ANOVA was conducted on residual scores as the dependent variable. No significant main effect was found for groups ( $F_{2,74}=2.85$ ,  $P=0.064$ ). Because of the operation of transforming raw scores into residual scores, testing for a main effect of scales was considered to be superfluous. A significant interaction effect was found ( $F_{4,146}=6.05$ ,  $P=0.000$ ; Wilks). Because of violation of the assumption of homogeneous variances (Levene's statistic =  $5.09$ ,  $P=0.008$ ), Dunnett's  $T_3$  test was used to compare mean VerBAS scores between aetiological groups. As multiple comparisons were in effect, the Bonferroni correction was conducted by dividing the nominal  $\alpha$ -level by the number of comparisons

with each single data set, resulting in a type I error level set at  $0.025$ . No significant differences were found between participants with DS and AS ( $P=0.058$ ), between participants with DS and PDD ( $P=0.967$ ), nor between participants with AS and PDD ( $P=0.179$ ).

Next, one-way ANOVA tests were used to compare groups in terms of their differential responding on the three scales of mand, tact and echoic, with raw residual scores as the dependent variable.

No significant difference of means on the mand scale was found between the three groups ( $F_{2,74}=1.36$ ,  $P=0.26$ ). Since no violation of the assumption of homogeneous variances occurred (Levene's test statistic =  $2.80$ ,  $P=0.067$ ), Tukey's HSD test was used. No significant differences were found between participants with DS and AS ( $P=0.277$ ), between participants with DS and PDD ( $P=0.413$ ), nor between participants with AS and PDD ( $P=0.966$ ).

There was no difference on the tact scale ( $F_{2,74}=3.19$ ,  $P=0.047$ ). Because of violation of the assumption of homogeneous variances (Levene's test statistic =  $7.79$ ,  $P=0.001$ ), Dunnett's  $T_3$  test was used. Testing revealed a nearly significant difference of means on the above scale between participants with DS and AS ( $P=0.027$ ), with means of  $2.09$  ( $SD=5.79$ ) and  $-1.55$  ( $SD=3.52$ ), respectively. No significant difference was found between participants with DS and PDD ( $P=0.332$ ), and between participants with AS and PDD ( $P=0.875$ ).

A one-way ANOVA revealed a significant difference of mean residual scores between the three groups for the echoic scale ( $F_{2,74}=10.12$ ,  $P=0.000$ ). Because of violation of the assumption of homogeneous variances (Levene's test statistic =  $7.31$ ,  $P=0.001$ ), Dunnett's  $T_3$  test was used. This analysis revealed a significant difference between means of residual scores of participants with DS and AS ( $P=0.000$ ), with means of  $1.42$  ( $SD=4.2$ ) and  $-2.78$  ( $SD=2.3$ ), respectively. No significant difference was found between participants with DS and PDD ( $P=1.000$ ). Finally, a significant difference in mean residuals was found between participants with AS and PDD ( $P=0.001$ ), with means of  $-2.78$  ( $SD=2.3$ ) and  $1.41$  ( $SD=4.71$ ), respectively.

## Discussion

The results of the present study suggest that the validity of the concept of behavioural phenotype is not refuted and that the concept of 'partial specificity' (Hodapp 1996) might be endorsed. There are several within-group differences. However, statistically significant differences between groups are minimal: individuals only differ in terms of their echoic communicative functioning. While those with AS perform better on manding than on tacting, those with DS show a reversed pattern.

It is surprising that individuals with PDD fail to differ in their verbal functioning. There is also no difference from those with DS. However, individuals with PDD may more often show delayed echoic responding, whereas those with DS more often show immediate echoic responses.

In examining their communicative competence, Mundy *et al.* (1988) found that, as compared to normal subjects matched for mental age, individuals with DS display a significant deficit in requesting for objects or requesting assistance in dealing with objects. This pattern was found to be specific for individuals with DS if compared with a sample of individuals with non-DS ID, a finding that is suggestive for the validity of the present finding. Concerning individuals with AS, the results obtained are consistent with those obtained by Penner *et al.* (1993) in that the above authors found that these individuals heavily rely on requests, being much like mands, while denying the use of comments or protest as a form of communication. The results obtained for the aetiological group with PDD are consonant with those obtained by Baron-Cohen (1989) and many others, suggesting that these children tend to have relatively better developed imperative communication skills than declarative communication skills, the latter being much like tacting.

The psychometric approach to behavioural phenotype research has the advantage in that it goes beyond easily observed traits or responses. However, the present study has several limitations. First, instrument insensitivity may have ignored highly unusual verbal responses that might characterize a given syndrome (e.g. delayed echolalia). Secondly, a general conclusion regarding the rela-

tionship between aetiology and communicative functioning requires the inclusion of individuals with other aetiologies. However, such an approach may have its limitations; for example, females with Rett syndrome are communicatively too different from males with fragile-X for any comparison.

Thirdly, participants may either have been similar or even more different in terms of their communicative functioning at a younger age, but evoke different responses from other people, ultimately leading to the communicative profiles which the present authors found. These are referred to as evocative genotype-phenotype interactions (Dykens 1995) and deserve further attention from researchers, who should focus on the extent to which parents, direct-care staff and teachers differentially react to children with a specific aetiology. Fourthly, the present authors relied on one type of measure only, i.e. the VerBAS. Other measures of communicative functioning could have been employed, such as the subscale of the VABS, although the psychometric properties of other instruments are often poor or unknown. Fifthly, participants often differed strongly in their modality of communication: those with DS and PDD most often used speech, whereas speech was not used by those with AS. Notwithstanding these differences, the VerBAS claims to estimate communicative functioning irrespective one's modality (see Duker 1999). Finally, one might argue that a correlation of 0.61 between ACB and VerBAS scores accounts for only 37% of the variance, thereby invalidating the operation to make groups comparable. While this may be true, justification of comparison of groups was also sought by employing strict inclusion criteria in using the VerBAS (see the section on 'Recording' above).

The results of the present study might be considered for their implications for intervention. Children with DS and PDD would probably profit from mand training since this aspect of communication is relatively underdeveloped with these groups. There are several empirically based procedures for mand training (e.g. Shafer 1994). However, because of their tendency to avoid social stimuli, it is likely to be more difficult to teach manding or requesting to individuals with PDD than those with DS. Tacting or labelling would be a training objective for those



with AS and, to a lesser extent, for those with PDD. The latter individuals will probably experience more difficulties because of impaired joint-attention skills.

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