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Differentiating the behavioural profile in autism and mental retardation and testing of a screener

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Introduction

Abstract In order to differentiate the behavioural profiles in autism and mental retardation and to cross-validate a behavioural autism screen, 84 subjects with autism (64 males and 20 females) with a mean age of 10 years selected from a Swiss national survey were compared to a control group of 84 subjects matched by age and gender with mental retardation, but without autistic features. The behavioural profile was assessed using the Developmental Behaviour Checklist (DBC). The behavioural profile in autism, in contrast to mental retardation, was marked by higher scores in the domains of

disruptive, self-absorbed, communication disturbed, anxious and autistic behaviour, and a higher total DBC score. Furthermore, a higher vulnerability for behavioural abnormalities became evident for females with autism. A recently proposed DBC-Autism Screen was cross-validated, and a slight extension of the screen led to even higher correct classification rates. It was concluded that the DBC is a suitable instrument for the assessment of the behavioural profile and for screening in autism.

Key words behavioural profile – autism – mental retardation

With its three core deficits in the areas of social deficits, communication, and restrictive and repetitive behaviour and interests, autism is a unique disorder that clearly distinguishes itself from all other mental disorders and, at the same time, applies to all individuals afflicted with autism. The behavioural phenomena have been clearly described in numerous studies, and there is a wide array of assessment devices depicting these behavioural features.

Currently, the most prominent structured interviews include the Autism Diagnostic Interview (ADI) [12] or the more recently developed Diagnostic Interview for Social and Communication Disorders (DISCO) [19]. In addition, structured observation schedules have been developed for clinical and research assessments like the Autism Diagnostic Observation Schedule (ADOS) [13] and its extension to children less than 6 years old, the Pre-linguistic Autism Diagnostic Observation Schedule [9]. On a third level, various questionnaires allow the study of behavioural phenomena in autism on a screening level at relatively low cost in terms of professional time that has to be invested. Examples include the Autism Screening Questionnaire (ASQ) [11], the checklist for Autism in Toddlers (CHAT) [1], or the Children's Social Behaviour Questionnaire (CSBQ) [14]. With the recent progress in the study of the genotype of autism, all these quantitative assessment devices may have become even more valuable when studying the relation between behavioural profile and genotype.

Given the considerable overlap between autism and mental retardation including the differential diagnosis between the two disorders, there is further need to clearly differentiate the behavioural features in the two types of disorders. More specifically, the study of genotype – phenotype relations needs to separately consider those aspects that are clearly autism-related and those that deal primarily with mental retardation. Consequently, the two disorders need to be studied by use of assessment devices that cover a wide range of behavioural phenomena of both clinical entities. The present study sought a differentiation of the behavioural profiles in a sample of autistic children and age- and gendermatched controls of mentally retarded children without autistic features by using a behaviour questionnaire which had specifically been designed for use in children with severe developmental disorders. In addition, a recently proposed screening algorithm from Australia (DBC-ASA) [2] was tested in terms of a replication and further validation.

Method

Samples

From a recent Swiss nationwide survey based on questionnaire responses by parents and caretakers of autistic individuals [17], we selected the total group of questionnaires dealing with autistic children and adolescents younger than 18 years. We identified 64 males and 20 females in this sample with a mean age of 10.72 (SD = 3.76) years. Since data collection was based on mailed questionnaires to be filled in by the members of the Swiss Autism Society, no professional check of diagnostic accuracy besides a questionnaire item asking for the diagnosis of autism could be made. According to the questionnaire, the sample comprised the following diagnoses: infantile autism (N = 30 or 36%), atypical autism (N = 30 or 36%), Asperger Syndrome (N = 2 or 2%), mental retardation with autistic features (N = 8 or 9%) and no information (N = 14 or 17%). In order to increase reliability of diagnosis, the survey questionnaire included the Autism Screening Questionnaire. In the final survey sample and in the present sample, data were retained only on those subjects who scored above the total ASQ score of 15, which has been established as a valid cut-off score for children with autism [11].

The control group was taken from another large national standardization sample of the Developmental Behaviour Checklist (DBC) (see below) that was recently collected in a random fashion by the authors with the help of the major German self-help group for mentally retarded individuals called *Lebenshilfe* (life assistance). Data were collected in various cooperating institutions across Germany. From this standardization sample, we took data of another 84 individuals matched by gender, age, and degree of disability to the autism sample. Children with a diagnosis of autism or autistic features in addition to mental retardation were excluded. The final control group included 64 males and 20 females with a mean age of 10.74 (SD = 3.75) years. Based on questionnaire information, the following diagnostic groups were obtained: Downs syndrome (N = 14 or 17%), other dysmorphic syndromes (N = 13 or 15%), cerebral palsy/epilepsy (N = 30 or 36%), brain damage/malformations (N = 6 or 7%), and non-specific etiologies (N = 21 or 25%).

The degree of disability was composed of a summary rating of four items, namely receptive language, active speech, self-care, and motor functioning. Each item was rated on a six-point scale ranging from 1 (very good functioning) to 6 (no function at all). Ratings were performed by the parents and caretakers. The two samples were well matched according to this disability rating with a mean of 3.67 (SD = 0.95) in the autism sample and a mean of 3.87 (SD = 1.00) in the mental retardation sample (t = 1.31, df = 166, p = n. s.).

Procedure

The study is based on responses to the Developmental Behaviour Checklist (DBC) by Einfeld and Tonge [3, 4]. This is a standardized instrument completed by lay informants to assess behavioural and emotional disturbance that was developed specifically for use with children and adolescents with mental retardation. It covers 96 behavioural items including two open questions that are rated on a three-point scale ranging from 0 (not true) to 1 (somewhat or sometimes true) and 2 (very true or often true). The DBC has good psychometric properties [3, 4, 8] and has also recently been used for the assessment of population prevalence of psychopathology of mentally retarded children and adolescents [5, 6].

Besides a total score, six subscales based on factor analyses can be computed. These subscales have been slightly revised based on new analyses by the authors [7]. The six subscales are labelled disruptive, self-absorbed, communication disturbance, anxiety, autistic relating, and antisocial. Because of a lack of a standardized scale for the DBC scores, we computed raw scores for the six subscales and transformed them into weighted raw scores (total subscore divided by the number of items of the respective subscale). This procedure allows a comparison of the various subscales among one another. In addition, a recently proposed DBC-Autism Screening Algorithm (DBC-ASA) [2] was calculated and tested.

Responders to the questionnaire were the parents and caretakers of the probands. As mentioned above, they also responded to the Autism Screening Questionnaire (ASQ) [11], which was used both for diagnostic certainty in the mailed survey and for tabulation of correlations with the DBC scale scores.

Statistical analyses

Group comparison was made using multivariate and univariate analyses of variance with gender as co-variate (MANCOVA and ANCOVA). In addition, Pearson correlation coefficients were computed. Finally, univariate logistic regression analyses were performed, and receiver operating characteristic curves (ROC) were plotted in order to test for the discriminative power of the DBC item. As in the study by Brereton et al. [2], a stringent criterion for significance was applied in logistic regression analysis in order to arrive at an effective diagnostic screen. All analyses were completed using the SPSS package.

Results

In a first step of the analyses, the behavioural profiles of the two samples were compared. The MANCOVA of the six subscale scores of the DBC resulted in highly significant differences for the two groups (WILKS Lambda = 0.69, F = 11.79, df = 6; 158, p < 0.001), whereas gender was not significant. As can be seen in Fig. 1, the subjects with autism scored significantly higher on all subscales except antisocial behaviour. Thus, children with autism displayed more disruptive, self-absorbed, communication disturbed, anxious, and autistic behaviour than the mentally retarded subjects without autistic features. Using these six subscales in a logistic regression analysis in order to predict group membership, it was found that the autistic subscale (B = 1.57, Wald = 5.16, p = 0.02) and the self-absorbed subscale (B = 2.62, Wald = 13.64, p = 0.0002) were the scales contributing significantly to a correct classification rate of 75.6% of all subjects.

As a consequence, the total DBC score was also significantly higher (F=36.13; df=1, p<0.001) in the subjects with autism (mean=62.20, SD=23.14) as compared to controls with mental retardation (mean= 43.20, SD=23.90). However, for the total score, there was a significant interaction of group by gender (F=8.21, df=1, p=0.005) as shown in Fig. 2, indicating that, in autism, females scored higher than males, whereas the reverse was true in mental retardation. Age was almost insignificant in both samples. The single exception was a relatively low, though significant, positive association of the DBC subscale measuring autistic behaviour in the autism sample (r=0.30, p<0.01).

The second goal of the study dealt with the discriminative power of the DBC. Findings from univariate logistic regression analyses are shown in Table 1. A comparison with the DBC–ASA shows that 21 out of the 29 items were clearly replicated at the p < 0.01 level and an additional three items were replicated at the p < 0.03level. Furthermore, two out of the five non-replicated items fell off very shortly with a p-value of 0.06 only. There were 11 additional items contributing to a significant differentiation of the two samples, including six items (Nos. 12, 17, 21, 26, 76, 85) that were also significantly associated with autism in the study by Brereton

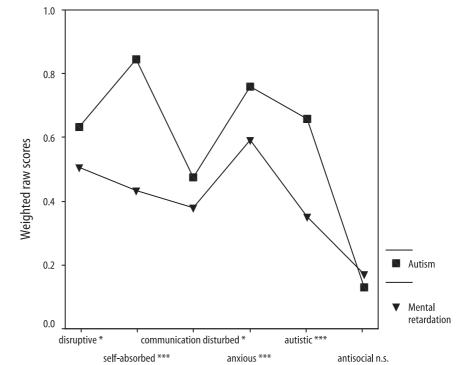
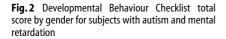
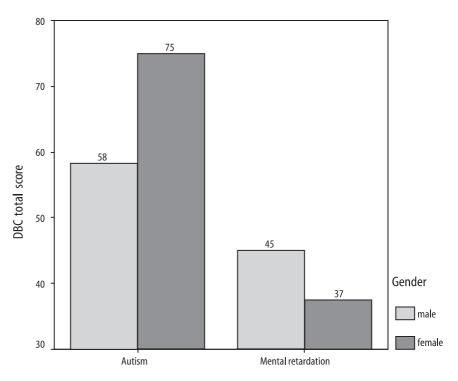


Fig. 1 Developmental Behaviour Checklist Profiles for subjects with autism and mental retardation * p < 0.05 ** p < 0.01 *** p < 0.001





et al. (2002), but not included in the DBC-ASA due to factorial loadings less than 0.70. In the present study, four of the other six items (Nos. 7, 33, 46, 72) are significantly more frequent in autism, whereas one single item (No. 20) is significantly less frequent in autism as compared to mental retardation.

In a further step, the original DBC-ASA and the revised DBC-ASAR1, as delineated from the present study including 40 items (29 original DBC-ASA items plus 11 newly identified items), were compared with regard to discriminative power. The area under the ROC curve (AUC) was 0.805 [95% confidence interval (CI) 0.74–0.87] for the DBC-ASA and 0.82 (CI 0.75–0.88) for the DBC-ASAR1. When deleting five non-discriminating items of the DBC-ASA and three items with itemtotal-correlation coefficients < 0.30 (Nos. 17, 20, 26), the AUC increased to 0.85 (0.79–0.90) for the DBC-ASAR2. Fig. 3 shows that the DBC-ASA algorithms and the two revised screens lead to curves including points with an acceptable trade-off between sensitivity and specificity.

A comparison of the sensitivity and specificity of the three scores as given in Table 2 shows that the original cut-off of 17 with an original sensitivity of 0.86 and a specificity of 0.69 in the study by Brereton et al. (2002) is not achieved in the present study in which the same sensitivity is obtained at a cut-off of 14 and a similar specificity is obtained at a cut-off of 17. The DBC-ASAR1 performs only slightly better in the present study and obtains the best cut-off at 21, leading to almost identical figures for sensitivity, whereas specificity is slightly worse than in the study by Brereton et al. [2]. Finally, at a cut-point of 17 the DBC-ASAR2 arrives at almost the same figures for both sensitivity and specificity as the DBC-ASA in the study of Brereton et al. (2002). The internal consistency coefficients (Alpha) for the three scores are also given in Table 2 and show a slight improvement of the coefficients for DBC-ASAR1 and DBC-ASAR2 as compared to the DBC-ASA. However, as Table 3 illustrates, the correct classification rate of the DBC-ASAR1 is slightly better than the DBC-ASAR2 and the DBC-ASA.

Discussion

The first goal of the present study was to identify a behavioural profile that may be considered to be specific to autism and may be differentiated from mental retardation as a common coexistent condition. Using the DBC as a suitable instrument for the collection of caregiver information and two age- and gender-matched samples of children with either autism or mental retardation without autistic features, the present study found a specific behavioural profile in autism with marked excesses on five out of six DBC subscales reflecting highly disruptive, self-absorbed, communication disturbed, anxious, and autistic behaviour. Even when using the recently proposed scoring revisions with a combined disruptive/antisocial scale [8], the differentiation between autism and mental retardation might not become less pronounced because the composition of the other five scales has not changed significantly. In the present

 Table 1
 Developmental Behaviour Checklist items differentiating autism from mental retardation without autistic features in univariate logistic regression analysis

No. Item	Wald $(df = 1)$	р
Replicated items of the DBC–ASA		
2. Avoids eye contact	5.94	0.015
3. Aloof, in his/her own world	30.54	< 0.001
5. Arranges objects routine in strict order	9.34	0.002
14. Deliberately runs away	4.84	0.03
18. Doesn't respond to others' feelings	21.18	< 0.001
25. Flicks, taps, twirls objects repeatedly	25.16	< 0.001
28. Obsessed with idea/activity	34.10	< 0.001
31. Has temper tantrums	9.96	0.002
34. Hums, whines, squeals, makes non-speech noises	33.88	< 0.001
35. Impatient	12.76	< 0.001
42. Laughs or giggles for no obvious reason	9.69	0.002
44. Likes to play with/hold unusual object, e.g. string	26.83	< 0.001
47. Mood changes rapidly for no apparent reason	15.09	< 0.001
50. Overactive, restless, unable to sit still	18.43	< 0.001
57. Prefers to do things on his/her own	28.80	< 0.001
58. Preoccupied with one or two interests	7.04	0.008
60. Repeated movements, e.g. handflapping/rocking	20.02	< 0.001
61. Resists being cuddled, touched/held	5.56	0.02
64. Smells, tastes/licks objects	9.28	0.002
66. Screams a lot	13.47	< 0.001
68. Stares at lights/spinning objects	14.82	< 0.001
86. Throws or breaks objects	10.59	0.001
89. Unrealistically happy/elated	7.09	0.008
94. Wanders aimlessly	17.16	< 0.001
Non-replicated items of the DBC-ASA		
9. Poor attention span	0.46	n.s.
43. Lights fires	1.00	n.s.
55. Poor sense of danger	0.24	n.s.
63. Repeats same word/phrase over	3.47	0.06
91. Upset over changes in routine/environment	3.50	0.06
Additional discriminating items	14.25	.0.001
7. Becomes over-excited	14.25	< 0.001
12. Covers ears/distressed by certain sounds	35.45	< 0.001
17. Doesn't show affection	7.75	0.005
20. Easily led by others	11.51	0.001
21. Eats non-food items, e. g. dirt, grass, soap	19.62	< 0.001
26. Fussy eater or has food fads	9.81	0.002
33. Hits self or bites self	11.56	< 0.001
46. Masturbates or exposes self in public	9.73	0.002
72. Switches lights on or off, pours water over and	25.39	< 0.001
over/similar repetitive activity	14.20	< 0.001
 76. Strips off clothes or throws away clothes 85. Tense, anxious, worried 	20.03	< 0.001
	20.03	< 0.001

study, it was decided to retain the original six-scale model in order to further allow comparisons with DBC data on specific mental retardation syndromes that were recently published by the authors [15, 16].

The insignificant age-effect on DBC scores, despite a wide age range between 3 and 17 years in both samples, supports the conclusion that the derived behavioural profiles are syndrome-specific and stable over time at least in childhood and adolescence. In contrast, the re-

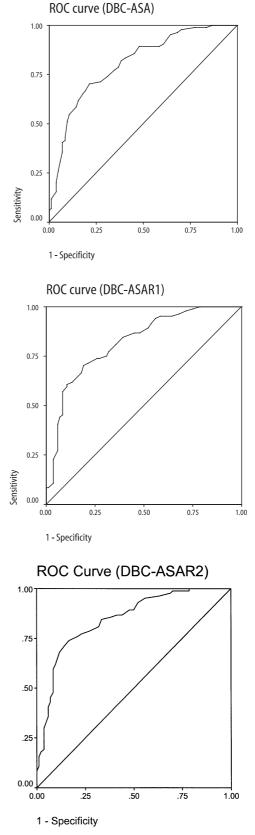


Fig. 3 Comparison of three screeners of autism based on ROC analysis

 Table 2
 Sensitivity, specificity, AUC, and reliability coefficients of three DBC screening algorithms for autism

Cut-off Point*	Sensitivity	Specificity	AUC	Alpha
DBC-ASA			0.80	0.91
14	0.86	0.55		
17	0.79	0.63		
DBC-ASAR1			0.82	0.93
21	0.85	0.61		
DBC-ASAR2			0.85	0.93
17	0.85	0.67		

* This value and above considered as cases; AUC = area under the ROC curve

 Table 3
 Classification of cases by three DBC screening algorithms for autism

		Percentag		
Algorithm	Nagelkerke R square	Autism	Mental retardation	Overall
DBC-ASA	0.62	85.7	79.8	82.7
DBC-ASAR1	0.67	88.1	86.9	87.5
DBC-ASAR2	0.58	82.1	86.9	84.5

markable gender-effect, with autistic girls showing more pronounced behavioural abnormalities than autistic boys, confirms the observation by Wing [19] that autistic girls, if affected by autism, display the more severe manifestation of the disorder which may be due to less genetic variability.

In the second part of the study, the diagnostic performance of the DBC was reconsidered after a screening algorithm had been introduced by Brereton et al. [2]. The comparison of the DBC-ASA as proposed by these authors and the present DBC-ASAR1 and DBC-ASAR2 arrived at very similar findings. First, the DBC-ASA was well replicated, given the fact that 24 out of 29 items were also identified for the DBC-ASAR1 and the DBC-ASAR2 and that ROC analyses resulted in very similar AUC rates. Thus, both studies show a similar screening performance to that of the ASQ [11]. However, the DBC covers a wider range of emotional and behavioural problems and has the advantage of indicating additional comorbid problems that are not addressed by screening instruments like the ASQ, the CHAT, or the CSBQ.

The slight differences in the diagnostic performance of the DBC-ASA, the DBC-ASAR1, and the DBC-ASAR2 with a slight superiority of the DBC-ASAR1 in terms of higher percentage of correct classification, a slightly higher reliability coefficient, and a slight superiority of the DBC-ASAR2 in terms of higher specificity may be sample-dependent. It must be kept in mind that the sample by Brereton et al. [2] was larger with an inclusion of 180 subjects in each of the two groups rather than 84 only in each group of the present study. Both the younger age of the Australian samples, compared with the present samples from Switzerland and Germany and culturally different approaches to management and service provision in the two studies may have contributed to the slight differences. The latter are by far less relevant than the consistency of the findings, pointing to the universal validity of the core features of autism.

While the use of the DBC-ASAR1 and DBC-ASAR2 may be advocated because of the inclusion of further items that are clinically, and not only statistically, significant for the diagnosis of autism, the final choice may be dependent on economic reasons like the different length of the three screening scales. Of course, one has to consider that the DBC-ASA as well as the DBC-ASAR1 and the DBC-ASAR2, with their limited specificity, lead to approximately 30% false positive rates at the proposed cut-off scores of 17 or 21, respectively. However, the three scales are proposed for screening rather than for the delineation of a clinical diagnosis.

Finally, some obvious limitations have to be addressed in the present study, as diagnosis in the two samples was not established by direct assessments of psychopathology and intelligence. In the sample of autistic subjects, the parents responded to mailed questionnaires, including various items describing the defining symptoms, the development of autism, and diagnosis. Thus, besides membership of a self-help group for individuals with autism, further detailed information on diagnostic status and the validated cut-off of 15 of the ASQ [11] were used to assure that only subjects with autism participated both in the larger survey and in the present study.

The control group of mentally retarded children and adolescents was also not directly assessed, but selected from a larger random sample that served for the German standardization of the DBC. All subjects were enrolled in intervention programmes and institutions for mentally retarded children and adolescents. Diagnostic information was obtained from the caregivers. The two groups were well matched with regard to degree of disability, compensating for the lack of direct IQ assessment. Given the general problems of proper IQ testing with increasing degree of disability, there is sufficient evidence that the two samples in the present study were of comparable developmental age.

Despite these limitations, there are some important clinical implications. The present study provides crossvalidated evidence from two culturally different regions of the world that the DBC is a useful device to screen children with developmental delays for autism. Given the distressing parental experience that diagnosis of autism is all too often delayed, as shown recently in the UK [10] and in the Swiss survey [17], the DBC might help in earlier diagnosis and onset of intervention.

The simple format of the DBC allows valid screening for autism and other emotional and behavioural symp-

toms in routine health screenings and in special populations of developmentally delayed children, so that referral for specialist assessment may be instituted as early as possible. Finally, the DBC may also be used clinically in order to evaluate the effects of specific interventions aiming at the reduction of emotional and behavioural problems in the large population of children with developmental disorders.

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