General Cognitive Abilities and Psychosocial Development in Children and Adolescents Having a Co-Twin with Down Syndrome

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Objective To examine the general cognitive and psychosocial development in children and adolescents having a co-twin with Down syndrome.

Study design A case control study with an individually matched control group was conducted. Participants included families with twins discordant for Down syndrome as well as with typically developing twins. The group of unaffected co-twins aged 4-16 years was compared with a control group of typically developing twins in terms of general cognitive abilities, behavioral problems, and prosocial behavior. The age and sex and the sex composition of the twins were individually matched. The Sijnders-Oomen nonverbal intelligence test was applied to assess children's IQ, and parents completed the Strength and Difficulties Questionnaire.

Results The unaffected co-twins did not differ from typically developing twins with respect to their IQ. Concerning the psychosocial development, significantly heightened values in unaffected co-twins twins were only obtained for the conduct problems scale (P = .01; r = 0.45), neither for the total difficulties score nor for the other behavioral problem scales significant differences were found.

Conclusions The general cognitive development of the unaffected co-twin of children with Down syndrome is not affected by the presence of their Down syndrome twin. Unaffected co-twins showed increased conduct problems, which is most pronounced in the younger children. *(J Pediatr 2021;232:214-9)*.

The psychosocial development, as well as the physical health, of children and adolescents who have a sibling with an intellectual disability, is strongly influenced by this experience.[1,2] Having a child with a disability such as Down syndrome presents all family members with a number of challenges, even more so when the constellation of dizygotic twins are discordant for Down syndrome. Owing to a number of sometimes severe health problems in the affected child, the load on the parents is high, which might result in a reduced amount of support for other members of the family.[3,4] The current study addressed the question of whether the cognitive and psychosocial development of the unaffected co-twin is affected by the presence of their Down syndrome twin.

In a study by Pilowsky et al, siblings of children with different types of disabilities (autism spectrum disorder [ASD], intellectual disability, developmental language delay) displayed general intellectual abilities within the average range.[5] Similarly, Olszewski et al did not find significant differences in full scale IQ between siblings of children with 22q11.2 deletion syndrome and community controls.[6] Also, no significant difference in nonverbal cognitive ability was reported for siblings of children with ASD compared with a group of siblings of typically developing children; however, the probability of speech-language difficulties was increased.[7] Conversely, Warren et al did not find significant group differences for language functioning nor for general cognitive ability between siblings of children with ASD and siblings of typically developing children.[8]

The psychosocial development of individuals with disabled siblings has been documented in numerous studies.[2] An increased risk of internalizing behavior such as anxiety and depression, but also various externalizing problems are described.[9-14] Higher levels of anxiety and depression were found for various groups of siblings of individuals with developmental disorders, except for siblings of individuals with Down syndrome.[15] There are also studies reporting no increased risk for behavioral or emotional problems in siblings of children with developmental disorders or positive outcomes.[16-22] The typically developing sibling's adjustments are determined by a number of factors, alongside the characteristics of the child with the disability (eg, the type and severity of the disability, the behavioral problems, the sibling relationship quality), and specific family characteristics (eg, the family's social attachment to supporting organizations, such as self-help groups).[2,19,23-25]

The specific challenges encountered by siblings of individuals with disabilities increase their risk to develop behavioral problems.[26] However, the generalizability of these findings is limited by methodologic problems, such as small sam-

ASD	Autism spectrum disorder
SDQ	Strength and Difficulties Questionnaire
SON-R	Snijders-Oomen nonverbal intelligence test

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Table I. Demographic information for the unaffected co-twins and the control groups

Variables	Unaffected co-twins (n = 31)	Control group (n = 31)	*P* value
Maternal age (years)	34.94 ± 4.85 (21-43)	33.54 ± 5.11 (23-49)	.30
Paternal age (years)	36.92 ± 5.52 (23-48)	38.15 ± 5.99 (29-57)	.58
Maternal education			.80
High school graduation or more	19 (61%)	17 (55%)	
Other	12 (39%)	14 (45%)	
Paternal education*			.61
High school graduation or more	15 (54%)	14 (45%)	
Other	13 (46%)	17 (55%)	
Gestational age (weeks)	34.97 ± 3.22 (27-40)	35.55 ± 2.72 (28-39)	.47
Other siblings (yes/no)	19/12	16/15	.61

Values are mean ± SD (range) or number (%), unless stated otherwise.
*There were missing values for 3 fathers in paternal education.

ple sizes, inappropriate grouping of samples with different types of disabilities, disregard of the socioeconomic status, and/or the absence of appropriate control samples.[2]

The present study evaluated whether in a group of twin pairs discordant for Down syndrome the general cognitive ability and/or the psychosocial development of the unaffected co-twin is influenced by the presence of his or her twin sibling with Down syndrome.

Methods

A case control study with an individually matched control group was conducted. The IQ, behavioral problems, and prosocial behavior were assessed in a group of unaffected co-twins aged 4-16 years. These factors were compared with a control group of typically developing twins individually matched in age and sex as well as the sex composition of the twins, while controlling for parental age and level of education and gestational age.

Participants

Participants were part of a comprehensive interdisciplinary study conducted at Saarland University in Saarbrücken and Homburg-Saar, Germany. The group of the unaffected co-twins consisted of 31 healthy dizygotic twin siblings (age range, 4-16 years; mean, 8.32 ± 3.67 years; 19 female), who had a co-twin with Down syndrome. At the time of the investigation, 28 families lived in Germany, 2 in Austria, and 2 in the German-speaking border region of France. The children and adolescents of the control group were matched on a case-by-case basis for sex and age as well as for the sex composition of the twins. In cases where the twins were of the same sex, the control twin was randomly selected. Accordingly, this group consisted of 31 typically developing dizygotic twins (age range, 4-16 years; mean, 8.33 ± 3.58 years; 19 female) who had a typically developing co-twin.

To control for further differences between the samples, paired sample *t* tests were run on various sociodemographic variables. No significant differences between groups appeared for the ages of the mother or the father at birth of the twins, nor for the week of delivery (all $P > .30$). Similarly, no significant difference between both groups was found for the maternal or paternal level of education or the number of siblings (χ^2 tests; $P > .60$). Accordingly, the 2 groups are well comparable with respect to sociodemographic characteristics (Table I).

All participants gave their written informed consent before their inclusion in the study. The present study was part of a research project, which was approved by the ethics committee of the Medical Association of the Saarland, Germany (protocol number: 195-08). For the recruitment of the families with twins discordant for Down syndrome, one of the authors, being a member of the advisory board of the German Down Syndrome Infocenter, informally contacted member families of this nationwide umbrella organization. In addition, pediatricians across Germany were contacted via the membership list of their professional society and calls for participation were published in several specialist journals and on the project's own homepage. The control group was recruited through calls in the regional press and study participant databases of several working groups of our university.

Measures

General Cognitive Ability. Participants completed the Snijders-Oomen nonverbal intelligence tests (SON-R 2.5-7, SON-R 5.5-17)[1].[27,28] For children younger than 6.5 years at the time of testing, the SON-R 2.5-7 was applied, and the older participants were tested with the short version of the SON-R 5.5-17. The latter encompasses the 4 subtests Categories, Mosaics, Analogies, and Situations that are also covered by the SON-R 2.5-7. For reasons of comparability, the analyses are based on these 4 subtests. Following the recommendation of the test authors, only the total IQ score is considered in this study and the specific scores obtained in the subtests are not compared separately.[27] Both tests can be performed within about 45-60 minutes.

Strength and Difficulties Questionnaire. The parent version of the Strength and Difficulties Questionnaire (SDQ)[29] is a screening questionnaire for the assessment of behavioral problems in children and adolescents ages 4-17 years. The 25 items of the German version are rated on a 3-point Likert scale (0-2) capturing 5 scales with 5 items

[1] The SON-R was chosen because this study is part of a comprehensive project in which the twins with Down syndrome were also tested.

each. Four of these encompass behavioral problems: the emotional symptoms scale, conduct problems scale, hyperactivity scale, and peer problems scale, which can be summarized in a total difficulties score. High values indicate problems in the respective domain. The fifth scale, prosocial behavior scale, captures positive behavioral aspects in social interaction with others with high values representing strengths in social competences.

Procedure

Families were visited at home by 2 trained experimenters. During these home visits, which took part either on 1 day or on 2 consecutive days, various developmental psychological testing procedures (including the SON-R, which was always applied first) were used. After a warming up phase, participants were tested with the SON-R, which lasted approximately 1 hour. Before the home visits, the families were sent consent forms and various questionnaires (including the sociodemographic questionnaire and the SDQ), which were collected on the day of the home visit. Children and adolescents were given a small gift and a certificate for participating.

Statistical Analyses

Using the procedure-specific evaluation software (SON-R, version 5.6), a standard IQ value (mean, 100 ± 15; min-max, 50-150) was determined on the basis of the performance shown in the subtests. The IQ* value used in the present study is based on this standard value, but corrects for the Flynn effect[30] by taking into account the period of time between the time when the normalization was established and the date of the test. For the SDQ, both, the total difficulties score as the sum of the 4 problem scales (range, 0-40) and the 5 subscale values (including the scale for prosocial behavior; range per scale 0-10) were calculated.

For data analyses, the statistics software package IBM SPSS Statistics 24 was used. The level for significance was set at an α of 0.05 and effect sizes were calculated using Cohen d for dependent-samples t tests. Because the participants in the control group were individually matched to the unaffected co-twins, dependent-samples t tests were calculated to compare the 2 groups. The distributions of the IQ* and the SDQ total difficulties score met the criteria for the use of parametric tests, whereas the subscales of the SDQ consisting of 5 items with 3-point Likert scales only allow for nonparametric tests owing to their skewness and low variance. Accordingly, Wilcoxon tests were applied here for testing the differences between groups. In addition, both the values of the SDQ total difficulties score and those of the SDQ subscales were assigned to the categories normal, borderline, and abnormal according to the cut-off values of the British norm cohort, with the categories borderline and abnormal being combined in the risk group.[31] The frequencies between the 2 groups were compared using χ^2 tests. Where analyses had expected cell counts were less than 5, results from the Fisher exact test are reported instead.

Results

General Cognitive Ability

Results indicate no significant difference in IQ* between the unaffected co-twins (mean, 106.4 ± 15.6) and the typically developing twins, mean, 102.7 ± 13.7; $t(30) = 1.00$; $P = .32$; $d = 0.18$. The general cognitive ability of the children and adolescents with a co-twin with Down syndrome is therefore not significantly different from that of individuals with a typically developing co-twin. To check whether the distribution of the IQ* values differs between the groups, a Kolmogorov-Smirnov Omnibus test was calculated for skewness and excess, which also was not significant ($P = .70$). One sample t tests, with which the IQ* values of the 2 group were tested for differences from the normative value (IQ of 100), showed a significant deviation from the normative value for the unaffected co-twins, $t(30) = 2.30$; $P = .03$; $d = 0.42$, but not for the control group, $t(30) = 1.12$; $P = .27$; $d = 0.20$.

Behavioral Problems

A t-test for dependent samples showed no significant difference in the SDQ total difficulties score between both groups, $t(30) = 0.63$; $P = .54$; $d = 0.10$ (Table II). Likewise, we found no significant difference in the distribution of the SDQ total difficulties score (Kolmogorov-Smirnov-Omnibus test; $P = .56$). Wilcoxon tests at the level of the SDQ subscales showed a significant difference between the 2 groups for the conduct problems score ($z = 2.49$; $P = .01$; $r = 0.45$), with the unaffected co-twins exhibiting higher average

Table II. SDQ mean scores, SDs, and number of participants assigned to the normal (N_n) or the risk (N_r) group for the unaffected co-twins, the control group and a normative sample

	Unaffected co-twins		Control group			Normative	
SDQ score	Mean ± SD	N_n/N_r	Mean ± SD	N_n/N_r	*P* value	Mean*	*P* value
Total difficulties score	7.84 ± 5.13	27/4	7.19 ± 3.71	30/1	.54	8.13	.75/.17
Emotional symptoms	1.81 ± 2.10	28/3	1.77 ± 1.50	26/5	.52	1.53	.22/.78
Conduct problems	2.45 ± 1.77	17/14	1.58 ± 1.21	28/3	.01	1.82	.06/.32
Hyperactivity	2.23 ± 2.06	30/1	2.61 ± 2.35	29/2	.43	3.19	.01/.13
Peer problems	1.35 ± 1.31	27/4	1.23 ± 1.06	27/4	.85	1.59	.04/.01
Prosocial behavior	7.58 ± 1.86	27/4	8.26 ± 1.75	29/2	.08	7.55	.74/.08

*No SDs were given in Woerner et al (2004).[32]

values than the twins of the control group. For the other problem scales, no significant group differences were found ($P > .43$). Similarly, no significant difference was obtained for prosocial behavior ($z = 1.75$; $P = .08$; $r = 0.32$). To determine if the behavioral problems are related to the age of the participants, correlational analyses were carried out for the SDQ total difficulties score as well as the subscale scores for each group separately. For the unaffected co-twins, significant correlations with age were obtained for the conduct problems score ($r = -0.36$; $P = .048$) and for the prosocial behavior score ($r = 0.47$; $P = .008$), indicating a reduction of conduct problems with increasing age as well as an increase in prosocial behavior with age. No significant correlations occurred for the control group ($P > .16$).

One sample *t* tests (total difficulties scores) and Wilcoxon tests (at the level of the SDQ subscales) were used to compare mean scores from the 2 groups with those from the normative data for the SDQ generated for German children and adolescents.[32] For the total difficulties score, no difference to the normative values was found for either group (P values $> .17$). Concerning the subscales, unaffected co-twins were rated as showing less peer problems ($z = -2.04$; $P = .04$) and less hyperactivity behaviors ($z = -2.60$; $P = .01$), and the rating in the other SDQ subscales did not differ significantly from the normative values (P values $> .06$). The control group also showed fewer peer problems ($z = -2.64$; $P = .01$), but did not differ significantly from the normative sample with respect to the other behavioral problem scales or the prosocial behavior scale (P-values $> .08$).

According to the cut-off values from the British norm cohort, participants were assigned to the categories normal, borderline, and abnormal, with the categories borderline and abnormal being combined in the risk group (**Table II**).[31] For the conduct problems scale significantly more unaffected co-twins were assigned to the risk group as compared with the typically developing twins (n = 14 vs n = 3; $\chi^2 = 7.12$; $P = .006$). For the other scales there were no significant differences ($P > .10$).

Discussion

The present study explored the general cognitive abilities and the psychosocial development in children and adolescents having a co-twin with Down syndrome. Unaffected co-twins did not differ from typically developing twins with respect to their general cognitive abilities, neither in their mean IQ value nor in the distribution of that value. In comparison with norms, unaffected co-twins exhibited a slightly increased IQ, whereas the typically developing twins did not differ from the normative value. With respect to psychosocial development, significantly heightened values in the unaffected co-twins were only obtained for the conduct problems scale, whereas for the other behavioral problem scales, no significant differences between the 2 groups were found. In comparison with norms, unaffected co-twins showed less hyperactivity behavior and fewer peer problems. Typically developing twins also received lower values on the peer problems scale.

It is important to note that the group of typically developing twins was comparable with the unaffected co-twins, because they were not only individually matched for the age and sex of the twin and for the sex composition of the twins, they also did not differ significantly with respect to important sociodemographic factors that have been shown to influence the cognitive and the psychosocial development of the children.[14,16]

In line with published findings, no difference in IQ between the 2 groups was found, thus, no disadvantages in the domain of general cognitive development can be discerned for the condition of having a co-twin with Down syndrome.[6,7] In contrast, a slightly increased IQ was noted in the twins as compared with normative data, which was not present in the control group. This small difference might be due to the small but insignificant differences in parental education between both groups, with parents of the twins discordant for Down syndrome having somewhat higher levels of education. In line with the results of the present study, it has been found that unaffected siblings of disabled children do not exhibit impairments in academic achievements as compared with controls, although they demonstrate a more negative attitude and more behavioral problems at school.[33,34]

With regard to the behavioral problems of children with a disabled sibling, meta-analyses have found an increased probability of psychosocial distress and behavioral problems.[22,25,35] In the present study, significantly increased values in the unaffected co-twins as compared the typically developing twins were only obtained for the conduct problems scale, whereas for the other behavioral problem scales, no significant differences between the 2 groups were found. With respect to the assignment to the categories normal vs risk group according to the cut-off values from the British norm cohort, most children and adolescents were assessed to be within the normal range by their parents (84% of the unaffected co-twins, 97% of the control group; total difficulties score).[31] The value for the unaffected co-twins is consistent with the one reported from the German representative sample (85%).[36] With respect to the subscales, the group of unaffected co-twins display more conduct problems compared with the control group, but lower values in hyperactivity compared with the normative value. A total of 45% of the unaffected co-twins were assigned to the risk group, as compared with 10% of the control group. In the German representative sample, the corresponding value amounts to 21%.[36]

Overall, the level of behavioral problems of the children and adolescents as reported by the parents is comparable between the 2 groups and as compared with normative values and far below all potential clinical limits. A small difference occurs with respect to conduct problems, which is partly in line with the results of a meta-analysis on siblings of children with chronic health conditions.[35] However, in addition to an

increased amount of externalizing behavior, the authors also report an increased probability of internalizing problems, which is not confirmed by the results of the present study. Siblings of individuals with ASD have significantly more negative outcomes overall as well as internalizing behavior problems, but no difference occurred with respect to externalizing behavior problems compared with the comparison groups.[22] Thus, the type of disability seems to be important for the probability that siblings of children with disabilities develop different kinds of behavioral problems.[37] Both groups were rated as having less peer problems compared with the normative value.

In the present study, for the group of the unaffected co-twins, a significant negative correlation with age was obtained for the conduct problems score and a significant positive correlation for the prosocial behavior score, whereas no such correlations were obtained for the control group. Thus, in the unaffected co-twins the conduct problems are decreased with advancing age and the amount of prosocial behavior increases. This finding indicates that differences from the control group are limited to the younger age and are reduced or even disappear with advancing age. Bailey et al found that in children with intellectual disabilities, behavioral problems, and prosocial behaviors improved with increasing age, whereas externalizing behavior problems were reduced during the 8-year period internalizing problems did not change systematically over time.[38] This finding suggests that the developmental changes observed in the unaffected co-twins in our study might be a result of the developmental changes in their co-twin with Down syndrome.

These findings must be considered in light of some limitations. First, the sample size is small and, thus, generalizations are limited. Furthermore, behavioral problems of the children and adolescents were rated by their parents. Parental reporting sometimes differs from children's self-reported behavior problems with siblings reporting greater behavioral problems than parents perceived them to have.[21] Because we were mainly interested in whether the group of unaffected co-twins differs with respect to behavior problems from the group of individually matched typically developing twins this problem might be of minor relevance because in both groups parents rated their child.

The present study shows that the majority of children and adolescents having a co-twin with Down syndrome are well-adjusted. Although these children and adolescents may experience challenges in their development while growing up with a co-twin with Down syndrome, the findings of the present study argue against the usually negative perspective on the situation of families raising a child with an intellectual and developmental disability. The evidence against that assumption presented here may well contribute to attenuating such doubts for the parents, and to achieve a well-informed decision within that multifaceted existential challenge. ■

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