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**Author:** Gameren-Oosterom, Helma van  
**Title:** Growth, development and social functioning of individuals with Down syndrome  
**Issue Date:** 2013-06-19
Problem behavior of individuals with Down syndrome, assessed in a nationwide cohort at late-adolescence

Authors
Helma B.M. van Gameren-Oosterom, MD
Minne Fekkes, PhD
Jacobus P. van Wouwe, MD, PhD
Symone B. Detmar, PhD
Anne Marie Oudesluys-Murphy, MB, PhD
Paul H. Verkerk, MD, PhD

Journal
Submitted

Chapter 8
Abstract

Objective: To assess problem behavior in adolescents with Down syndrome (DS) and study the relation to gender and severity of intellectual disability.

Study design: Cross-sectional data of a Dutch nationwide cohort of DS children, aged 16-19 year, were collected using a written parental questionnaire. Problem behavior was measured using the Child Behavior Checklist (CBCL) and compared to normative data. The degree of intellectual disability was determined using the Dutch Social competence rating scale (SRZ). Differences were evaluated using t-tests and linear regression analysis.

Results: Response was 62.8% (322/513), mean age 18.3 years (SD ± 0.8). Total score of problem behavior was higher in adolescents with DS compared with adolescents without DS (26.8 vs. 16.5, p<0.001). Overall, 51% of adolescents with DS had problem scores within the clinical or border range on one or more CBCL subscales; more than twice as high as adolescents without DS. Adolescents with DS showed more internalizing problems (14% vs. 9% within the clinical range) and externalizing problems were almost equal (7% vs. 9% within the clinical range). Highest problem scores were observed on the subscales social problems and thought problems (with large to very large standardized differences). Male gender and/or more severe mental retarded were associated with more behavioral problems.

Conclusions: Serious problem behavior is highly prevalent in adolescents with DS. This demonstrates the need for attention for general behavior improvement as well as for detection and treatment of specific psychopathology in individuals with DS.
Introduction
Down syndrome (DS), Trisomy 21, is the most prevalent cause of intellectual impairment. In the United States the prevalence of DS is estimated to be 12 per 10,000 live births; in the Netherlands 14.6 per 10,000 live births (annually approximately 245 children with DS are live born).\(^1,2\) Children with DS have delayed cognitive and motor development as well as specific medical problems, e.g. congenital heart defects, gastro-intestinal disorders, thyroid dysfunction and visual impairment.\(^3,4\) Moreover, it is known that children with DS are prone to psychopathology; prevalence estimates range from 18% to 38%.\(^5-8\) This risk is lower than in other forms of intellectual disability.\(^5\)

The patterns of problem behavior in children change with age, especially during adolescence since this period is characterized by changes, hormonally, physically, psychologically and socially.\(^9\) Adolescents with DS also have to deal with puberty, sexual development, (start of) emotional separation from parents and development of social autonomy.\(^4,10\) Some studies confirmed that also among children with DS changes in behavioral pattern occur at adolescence, i.e. externalizing symptoms decreased whereas internalizing symptoms increased.\(^11,12\)

The few studies on behavioral problems in adolescents with DS are limited as are studies on the ‘dual diagnosis’ of intellectual disability and psychopathology, mainly because of the small sample sizes (<60) and broad age ranges (mostly 4-19 years).\(^7,8,13,14\) No large sample studies describing behavior in DS at a late teen age could be found. Also, gender effects in relation to behavioral problems are barely reported in DS, while these are well known in the general population.\(^11,15\) This study aims to examine problem behavior at late adolescence in a large nationwide cohort of individuals with DS and its relation to gender and the degree of intellectual disability.

Patients and Methods

Participants
Data were collected from a nationwide Dutch cohort of parents of children with DS, assessed at the age of 16-19 years. This cohort included children with DS born in 1992, 1993 and 1994. Of all children with DS born in those 3-years period, an estimated 595 adolescents were still living in the Netherlands (based on an 81% survival rate).\(^2,16\) The Dutch Down Syndrome Foundation (parent organization) had contact with 86% of these parents and sent them a written request. The only selection criterion for inviting parents to participate was the year of birth of the DS child. Parents could respond within 4 months after receiving the invitation. Reminders were sent after 4 and 8 weeks.
Measurements and procedure

Parents completed a written questionnaire consisting of two validated tests and additional questions on background and level of functioning. Written informed consent was obtained from parents/next of kin of all participants.

The Dutch version of the Child Behavior Checklist (CBCL) for 4-18-year-old children was used to measure problem behavior. Although the CBCL has been developed for children with normal intelligence, it is frequently reported to be suitable for children with developmental delay. Normative data are available in the test manual for the age group of 12-18-year-old adolescents. Additionally, normative data on mean scale scores of the CBCL were available from 15-18-year-old adolescents in the general Dutch population as published by Bongers. Both normative data were based on parental report. Because Bongers’ sample resembles ours mostly, these normative data were used for comparing mean scale scores and normative data from the test manual for the other comparisons.

The CBCL contains 113 problem behavior items rated from 0 (not true) to 2 (very true or often true). A total problem score can be calculated using these items. The items of the CBCL can also be grouped into the following eight subscales: withdrawn, somatic complaints, anxious/depressed, social problems, thought problems, attention problems, delinquent behavior and aggressive behavior. A scale of internalizing problems is constructed by combining the subscales withdrawn, somatic complaints and anxious/depressed. The scale externalizing problem is formed by combining the subscales delinquent behavior and aggressive behavior. Moreover, all scale scores can be grouped into scores within the normal, border or clinical range of the scale.

The Dutch Social competence rating scale (SRZ) was used to determine the degree of intellectual disability. This validated instrument measures social independence specifically in mentally disabled children aged 4-18 years and has already been established as a sensitive instrument to measure changes in self-help skills in adults with DS. The SRZ was selected for its ability to measure intelligence quotient scores in the lower zones of the scale, whereas some other intelligence tests – such as the Wechsler Intelligence Scale for Children (WISC) – are not. The SRZ contains 31 items that measure skills needed for independent functioning in daily life. Based on these items, the degree of intellectual disability (mild, moderate, severe or profound) can be determined. These degrees represent the following self-help skills:

- **Profound intellectual disability** means hardly able to dress oneself, wash hands and face properly and use adequate toilet hygiene, just able to eat independently (without the use of a knife) and barely able to speak.
- **Severe intellectual disability** means able to undress, wash hands and face, use a knife and fork at dinner, clear up after dinner, speak using incomplete sentences with unclear
pronunciation and can be understood only by close caregivers or familiar people.

- **Moderate intellectual disability** means the adolescent dresses himself completely, washes hands and face properly, uses adequate toilet hygiene, uses a knife and fork at dinner including cutting meat (without a bone), able to walk outside the home without supervision and his speech can mostly be understood by others.

- **Mild intellectual disability** means able to dress oneself completely including footwear, maintain complete personal hygiene, set the table properly, walk about several streets away from the home without supervision, use full or more compound sentences when speaking and speech and language can be understood by most others.

**Statistical analyses**

General characteristics of the study population were determined and compared between DS boys and girls using *t*-tests for continuous variables and chi-square tests for categorical variables.

Mean raw CBCL scale scores of boys and girls with DS were compared to normative data from 15-18-year-old adolescents in the general Dutch population as published by Bongers.\(^{15}\)

To evaluate the differences between mean values, *t*-tests were used and the standardized differences were estimated by dividing the differences in mean scores between the subgroups by the pooled standard deviation (SD). Cohen’s standardized differences \((d)\) were used for interpretation of relevant differences: \(d<0.2\) is considered a negligible difference, \(0.2\leq d<0.5\) a small, \(0.5\leq d<0.8\) a moderate, \(0.8\leq d<1.3\) a large and \(d\geq 1.3\) a very large difference.\(^{24}\)

Linear regression analysis was performed to assess the association between intellectual disability and the total CBCL problem score, adjusting for parental education and gender. In addition, to determine whether the effect of gender on the outcome variable was equal for all degrees of intellectual disability, the influence of interaction terms was assessed by linear regression analysis. For this purpose, cross products were computed between degree of intellectual disability and gender. These cross products were added as an extra step to the regression equation (which included all main effects).

For all analyses, statistical tests were 2-tailed and statistical significance was defined at *p*<0.05. The analyses were performed using Statistical Package for the Social Sciences, version 20.0 for Windows (SPSS Inc, Chicago, Illinois).

**Results**

In total, 322 of 513 sent questionnaires (63%) were completed. The mean age of the 322 participants was 18.3 years (SD=0.82, range 16.8-19.9 years) and 53% were boys. Ten per cent of adolescents were (very) profoundly mentally retarded, 30% severely, 43% moderately and 17% mildly. More boys with DS than girls scored a severe or profound
intellectual disability ($p<0.001$) and less boys than girls were mildly mentally retarded ($p=0.004$). Table 8.1 shows the general characteristics of our study sample.

Table 8.1: Characteristics of the study population of adolescents with Down syndrome ($n=322$), as reported by their parents; grouped by gender.

<table>
<thead>
<tr>
<th>General characteristics</th>
<th>Total</th>
<th>Boys</th>
<th>Girls</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number of subjects</td>
<td>322</td>
<td>100.0</td>
<td>170</td>
</tr>
<tr>
<td>Age in years (range)</td>
<td>16.8 - 19.9</td>
<td>16.9 - 19.9</td>
<td>16.8 - 19.8</td>
</tr>
<tr>
<td>Age in years (mean ± SD)</td>
<td>18.32 ± 0.82</td>
<td>18.34 ± 0.82</td>
<td>18.29 ± 0.82</td>
</tr>
<tr>
<td>Dutch descent*</td>
<td>300</td>
<td>93.2</td>
<td>162</td>
</tr>
<tr>
<td>Living at home</td>
<td>283</td>
<td>87.9</td>
<td>149</td>
</tr>
<tr>
<td>Parental education</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Low</td>
<td>39</td>
<td>12.1</td>
<td>23</td>
</tr>
<tr>
<td>Middle</td>
<td>105</td>
<td>32.7</td>
<td>58</td>
</tr>
<tr>
<td>High</td>
<td>177</td>
<td>55.1</td>
<td>89</td>
</tr>
<tr>
<td>Level of mental disability</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mild</td>
<td>54</td>
<td>16.8</td>
<td>16</td>
</tr>
<tr>
<td>Moderate</td>
<td>139</td>
<td>43.3</td>
<td>73</td>
</tr>
<tr>
<td>Severe</td>
<td>97</td>
<td>30.2</td>
<td>58</td>
</tr>
<tr>
<td>Profound</td>
<td>31</td>
<td>9.7</td>
<td>23</td>
</tr>
</tbody>
</table>

Abbreviation: SD – standard deviation,
*Boys with Down syndrome compared to girls with Down syndrome
* Both parents born in the Netherlands

Problem behavior

The total problem score of behavior as measured by the CBCL was higher in adolescents with DS (mean score of 26.8) in comparison to the normative sample of 15-18-year-old adolescents without DS (mean score of 16.5); a moderate standardized difference was found. Table 8.2 shows all mean scores of the test scales, where higher scores denote more problems. Grouping the total problem score into the normal or clinical range of the scale showed that 21% of adolescents with DS had a total problem score within the clinical range, compared to 9% in the normative sample (12-18-year-old adolescents without DS), see Figure 8.1. Overall, 51% of adolescents with DS scored in the clinical range of one or more subscales; more than twice that for adolescents without DS.

Both boys and girls with DS showed more internalizing problems than their peers, with a small standardized difference. Fourteen percent of adolescents with DS had a score within the clinical range vs. 9% in the normative sample. This was shown by problems on the subscales withdrawn and somatic complaints, whereas less problems in the subscale
Figure 8.1: Proportion of boys and girls with Down syndrome (aged 16-19 years; n=317) with behavioral problem scores within the clinical or border area, as measured by the Child Behavior Checklist; compared to a normative sample of 12-18-year-olds without DS.

Anxious/depressed were observed in the total DS sample and in DS girls, compared to the norms. No statistically significant difference was observed on the externalizing problem scale.

The largest standardized differences were found for boys and girls with DS on the subscales social problems, thought problems and attention problems (all three not grouped within the externalizing or internalizing problems scales). Here very large standardized differences were observed on social problems in boys as well as in girls and on thought problems in boys. In detail, this concerns the following problems: the subscale social problems mainly concerns problems with age appropriate behavior, clumsy coordination and being too dependent on adults; the scale thought problems concerns problems with obsessive thoughts, repetitive acts and weird behavior; the scale attention problems involves problems with concentration, being too active, impulsiveness and nervousness. When scale scores were categorized in clinical scores, up to 40% of adolescents score within the clinical...
Table 8.2: Problem behavior of 16-19-year-old people with Down syndrome (n=317), measured by the Child Behavior Checklist; grouped by gender and compared to 15-18-year-olds without Down syndrome; higher scores denote more problems.

<table>
<thead>
<tr>
<th></th>
<th>Total DS sample</th>
<th>Male</th>
<th>Female</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>(n=317)</td>
<td>DS (n=166)</td>
<td>Norm (n=1016)</td>
</tr>
<tr>
<td><strong>Total problems</strong></td>
<td>26.76 (15.85)</td>
<td>28.38 (16.68)</td>
<td>16.52 (15.14)</td>
</tr>
<tr>
<td><strong>Internalizing problems</strong></td>
<td>7.09 (5.39)</td>
<td>6.85 (4.90)</td>
<td>4.78 (5.15)</td>
</tr>
<tr>
<td>Withdrawn</td>
<td>3.65 (2.95)</td>
<td>3.52 (2.67)</td>
<td>2.12 (2.29)</td>
</tr>
<tr>
<td>Somatic complaints</td>
<td>1.73 (1.99)</td>
<td>1.71 (1.81)</td>
<td>0.76 (1.36)</td>
</tr>
<tr>
<td>Anxious/Depressed</td>
<td>1.82 (2.38)</td>
<td>1.73 (2.24)</td>
<td>1.97 (2.84)</td>
</tr>
<tr>
<td><strong>Externalizing problems</strong></td>
<td>5.02 (4.78)</td>
<td>5.81 (5.05)</td>
<td>5.86 (6.52)</td>
</tr>
<tr>
<td>Delinquent behavior</td>
<td>1.20 (1.52)</td>
<td>1.40 (1.59)</td>
<td>1.34 (2.10)</td>
</tr>
<tr>
<td>Aggressive behavior</td>
<td>3.82 (3.76)</td>
<td>4.41 (4.05)</td>
<td>4.52 (4.98)</td>
</tr>
<tr>
<td><strong>Social problems</strong></td>
<td>4.07 (2.15)</td>
<td>4.01 (2.15)</td>
<td>1.04 (1.72)</td>
</tr>
<tr>
<td><strong>Thought problems</strong></td>
<td>1.44 (1.85)</td>
<td>1.70 (2.05)</td>
<td>0.25 (0.73)</td>
</tr>
<tr>
<td><strong>Attention problems</strong></td>
<td>5.86 (3.41)</td>
<td>6.53 (3.84)</td>
<td>3.12 (3.18)</td>
</tr>
</tbody>
</table>

* p<0.05, ** p<0.01, *** p<0.001

Norm population, published by Bongers et al, 2003

Cohen’s effect size: d<0.2 negligible; 0.2≤d<0.5 small; 0.5≤d<0.8 moderate; 0.8≤d<1.3 large; d≥1.3 very large

Combined from the subscales withdrawn, somatic complaints and anxious/depressed

Combined from the subscales delinquent and aggressive behavior

Abbreviations: DS – Down syndrome, SD – standard deviation, NS – not statistically significant
Problem behavior of adolescents with Down syndrome

or border ranges of the subscale social problems, compared to about 6% of adolescents without DS.

Gender differences

Boys showed somewhat more problem behavior than girls within the DS sample. The difference was statistically significant for the externalizing problem scale ($p=0.002$) and on the subscales thought problems, attention problems, delinquent behavior and aggressive behavior ($p$-values 0.008, <0.001, 0.012 and 0.003, respectively). However, no overall difference is observed in the proportion of boys and girls with DS scoring in the clinical range of one or more subscales (52% and 50%, respectively).

On the subscale social problems no difference on mean scales score was observed between boys and girls DS, however more boys (24%) than girls (13%) scored within the clinical area of this subscale ($p=0.019$). When the proportion scoring within the border area (where the proportion of girls is larger) is added, this gender difference disappeared.

In the norm population (15-18-year-old adolescents without DS) a gender difference is also noticed. Although on the total problem score no differences were observed between boys and girls without DS, they were observed on externalizing problems and internalizing problems. Boys without DS also scored more externalizing problems than girls without DS, analogous to the observed gender difference within our DS sample. Conversely, higher problem scores were observed in girls without DS than in boys without DS on internalizing problems; this gender difference, (unfavorable for girls) was not observed among adolescents with DS.

Problem behavior and degree of intellectual disability

A statistically significant association was observed in adolescents with DS between the degree of intellectual disability and problem behavior (Table 8.3). The total problem score increased with the severity of intellectual disability. Adolescents with mild intellectual disability experienced the least problems (mean total problem score 19.7) and those with profound intellectual disability the most (mean total problem score 41.7). The interaction term on gender and degree of intellectual disability was not statistically significant ($p=0.057$).

Discussion

In a nationwide cohort of 322 Dutch adolescents with DS between the ages of 16-19 years, problem behavior was found to be more severe than appropriate for their age. Overall, half of the adolescents with DS had problems within the clinical or border range on one or more subscales; more than twice as high as adolescents without DS. The problems were most pronounced on the subscales social problems and thought problems. Boys with DS showed more behavioral problems than girls with DS on externalizing problems, as do boys without
Table 8.3: Linear regression analysis of the total problem score of the Child Behavior Checklist (CBCL) in 16-19-year-olds with Down syndrome (n=317); negative β means less behavioral problems compared to adolescents with moderate intellectual disability (reference group) and positive β more.

<table>
<thead>
<tr>
<th>Degree of intellectual disability</th>
<th>Unadjusted β</th>
<th>95% CI</th>
<th>Adjusted β†</th>
<th>95% CI</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mild</td>
<td>- 4.67</td>
<td>[-0.01; -9.33]</td>
<td>- 4.92</td>
<td>[-0.18; -9.67]</td>
</tr>
<tr>
<td>Moderate (reference)</td>
<td>0</td>
<td>0</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Severe</td>
<td>5.01</td>
<td>[8.89; 1.12]</td>
<td>4.66</td>
<td>[8.60; 0.72]</td>
</tr>
<tr>
<td>Profound</td>
<td>17.30</td>
<td>[23.07; 11.53]</td>
<td>17.13</td>
<td>[22.94; 11.31]</td>
</tr>
</tbody>
</table>

*Degree of intellectual disability is based on all items of the Dutch Social competence rating scale (SRZ)
† Unstandardized regression coefficient, adjusted for parental education and gender

DS in comparison to girls without DS. An association between the degree of intellectual disability and behavior was observed even after adjusting for parental education and gender: adolescents with DS with more severe intellectual disability were prone to have more behavioral problems.

Our results show that internalizing problems are more severe during adolescence while externalizing problems are not prominent in individuals with DS; this is in line with other studies.11,12 Myers and Pueschel studied a sample of 261 individuals with DS under 20 years of age (mean: 9.5, range 1-19) and observed that they often showed disruptive behavior, anxiety disorders and repetitive behavior.7 It has to be considered that this study was performed more than 20 years ago. Compared to their results, we also found more disruptive and repetitive behavior, however, a statistical significantly lower problem score was observed on the subscale anxious/depressed of the CBCL. Contrary to our result of lower depression scores in DS, other studies among adults with DS reported prevalence rates of depression ranging from 0 to 11%.25 Estimates rates of depression in adolescents are not available to our knowledge.

The individuals with DS in this study have previously been assessed at the age of 8 year.26 At that age, the children also showed more problem behavior than appropriate for their age. This was most pronounced on the subscales social problems, thought problems and attention problems, identical to the findings in this study at the age of 16-19 years. They experienced fewer problems on the subscale anxious/depressed at the age of 8 as well as at 16-19 years. The proportions of children/adolescents scoring within the clinical area of the total problem scale of the CBCL were comparable in both assessments: 27% at 8 year (2.5 times more than in the normative sample) and 21% at 16-19 year (2.3 times more than in the normative sample). Also the same observed gender differences in problem behavior were found at the ages of 8 and 16-19 years.
Our results indicated that more severe intellectual disability was associated with more problem behavior. This association has not been previously described in DS populations. Bongers et al. showed that the mean score of the subscale social problems will decline with age (highest score of 1.5 at 9 years and lowest score of 0.8 at 18 years) in the general population, suggesting that the higher score in our DS sample is caused by their lower developmental age.\(^1\) However, this does not adequately explain the observed differences, since the mean scale score was 4.1 in adolescents with DS. This means that, adolescents with DS have more social problems, regardless of age. This also applies to the subscales withdrawn, somatic complaints, thought problems and attention problems.

The present study includes a substantial part (54%) of all adolescents with DS living in the Netherlands and born in the included 3-years birth period. An additional strength is the wide spectrum of behavioral problems and background variables that were measured. This study presented the opportunity to investigate the association between problem behavior and gender and of degree of intellectual disability. However, some potential limitations should be noted. There may be selection bias in our study. The participants were invited by the parent organization. It is possible that parents with more positive attitudes may be more inclined to join such an organization and to participate in our study. However it is also possible that other parents with more concerns about their child may be more inclined to do so. Furthermore, our results are based on parental report. Parents may be tempted to emphasize positive aspects of their child’s behavior. However, also the normative data were based on parental report. Despite of the potential positive information bias, the results of our study indicate that a large range of clinically important behavioral problems are present in adolescents with DS.

**Implications**

This study demonstrates the extent of overall problem behavior in adolescents with DS. Regrettably we found no support for the stereotypical perception of children with DS as being charming, friendly and joyful individuals. In our study we observed that adolescents with DS are happy and not anxious or worried and generally not aggressive or delinquent. At the same time, our results indicate that many adolescents with DS are withdrawn, have large social problems and thought problems. The combination of these characteristics results in a perception for others, that adolescents with DS are in general compliant with others (happy, not aggressive and not worried), but they experience many problems when they have to stand up for themselves. Therefore, the stereotypical image of individuals with DS, held by many people including professionals, fails in recognizing the great extent of problem behavior that limits adolescents with DS in their daily functioning. Professionals, parents and all others need to be informed about these characteristics of adolescents with DS, so they can socially interact in an appropriate manner and evaluate their capabilities suitably.
In consequence, special care needs to focus on general improvement of behavior in children and adolescents with DS as well as on detection and treatment of psychopathology in individuals. With regard to the first point, it may be desirable to have a structured intervention program which focuses on training social and behavioral skills. At present, early intervention programs are generally available for only young children (up to the age of 6 years). When such intervention programs for older children and adolescents are developed and evaluated, these may provide parents and professionals with essential tools for stimulating and improving behavior of individuals with DS. To be able to develop optimal tools, it is first necessary to investigate to what extent behavior can be improved and how problem behavior can be prevented. The other point of interest, the ‘dual diagnosis’ of intellectual disability and psychopathology, has not been studied much in adolescents with DS. Current prevalence estimates of neurobehavioral and psychiatric co-morbidity in children with DS vary from 18% to 38%.\(^6\,^8\) Since limited social skills are a characteristic aspect of both intellectual disabilities and pervasive developmental disorders, definitions of ‘dual diagnosis’ are complex.\(^27\) Therefore, appropriate definitions of psychopathology in DS (such as attention deficit and hyperactivity disorders and autism) are needed, as well as screening instruments to detect these. Furthermore, treatment of specific behavioral disorders in individuals with DS needs to be improved.

**Conclusions**

Serious problem behavior is highly prevalent in adolescents with DS. Our findings emphasize the need for prevention, detection and treatment of these behavioral problems in adolescents with DS. Professionals need to be alert to the increased risk of behavioral problems. Furthermore, (expectant) parents of a child with DS have a right to information and counseling concerning the increased chance that their child will have behavioral problems.

**Acknowledgement**

We gratefully thank all parents who participated in the present study for their willingness and efforts to fill in the questionnaires. We also thank the Dutch Down Syndrome Foundation for inviting parents to participate in the study.

The study is financed by the Nederlandse Stichting voor het Gehandicapte Kind (NSGK, Dutch foundation for disabled children). The funders had no role in study design, data collection, analysis and interpretation of data, decision to publish, or preparation of the manuscript.
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