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## **Distress and everyday problems in Dutch mothers and fathers of young adolescents with Down syndrome**

Hedy A. van Oers<sup>a,1</sup>, Jan Pieter Marchal<sup>a,b,1</sup>, Heleen Maurice-Stam<sup>a</sup>,  
Martha A. Grootenhuys<sup>a</sup>, A.S. Paul van Trotsenburg<sup>b,2</sup>, Lotte Haverman<sup>a,2,\*</sup>

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a Academic Medical Center, University of Amsterdam, Psychosocial Department, Emma Children's Hospital, Amsterdam Public Health research institute, Post Box 22660, 1100 DD, Amsterdam, The Netherlands

b Academic Medical Center, University of Amsterdam, Department of Paediatric Endocrinology, Emma Children's Hospital, Amsterdam Public Health research institute, Post Box 22660, 1100 DD, Amsterdam, The Netherlands.

\* Corresponding author at: Academic Medical Center, University of Amsterdam, Psychosocial Department, Emma Children's Hospital, Amsterdam Public Health research institute, Post Box 22660, Room G8-136, 1100 DD, Amsterdam, The Netherlands.

1 Shared first author.

2 Shared last author.

## **Abstract**

### **Background**

To provide targeted support to parents of children with DS, knowledge of their distress and everyday problems is crucial. For this purpose, psychosocial screening instruments can be a valuable addition to routine clinical practice.

### **Aims**

To determine differences on a psychosocial screener concerning distress and everyday problems in parents of young adolescents (YAs) with DS versus control parents and in mothers of YAs with DS versus fathers.

### **Methods and Procedures**

We compared outcomes of the Distress Thermometer for Parents in 76 mothers and 44 fathers of 11 to 13-year-olds with DS versus 64 mothers and 52 fathers of age-matched children without DS (comparing mothers and fathers separately). Additionally, we compared mothers and fathers within 34 parent couples of YAs with DS.

### **Outcomes and Results**

Clinical distress was not more frequent than in control parents. Mothers further did not report more everyday problems and only differed from their controls on one problem domain and some problem items. Fathers, however, reported more problems than their controls across most domains and wished to talk to a professional about their situation more frequently. Outcomes in mothers and fathers within parent couples did not differ significantly.

### **Conclusions and Implications**

This is one of few studies to report on the use of psychosocial screening instruments in parents of children with DS. Our results suggested that attention for fathers of YAs with DS is required. Psychosocial screening instruments that inquire about specific problems and the wish for referral can play an important role in achieving this.

### **Keywords**

Down syndrome, parents, fathers, distress, everyday problems

### **What this paper adds**

Although numerous studies have focused on parental distress, this paper is one of few to systematically assess everyday problems in parents of young adolescents with DS. Moreover, this paper incorporates the perspectives of both mothers and fathers. We used a psychosocial screening instrument that is frequently used in clinical practice: the Distress Thermometer for Parents. Neither mothers nor fathers of young adolescents with DS reported more clinical distress than control mothers and fathers. Furthermore, mothers only differed slightly from control mothers in the frequency of reported problems. Fathers, however, reported significantly more everyday problems than their controls across a wide range of domains and showed more interest in talking to a professional about their situation. These relatively unfavourable outcomes in fathers suggest that their needs should be attended to in the care for families of young adolescents with DS. The distress score alone did not reveal these needs, while the problem items did; this suggests that discussion of specific problems and the wish for referral is required in clinical practice to determine the need for assistance. Screening instruments such as the one used in this paper can facilitate a targeted discussion of these psychosocial issues.

## **Introduction**

### **Background**

Down syndrome (DS) is the most common genetic cause of intellectual disability (De Graaf, et al., 2011). Besides intellectual disability, DS is accompanied by increased risks for health problems such as congenital heart disease, endocrine disorders, and leukaemia (Roizen & Patterson, 2003). Parents of children with DS have been generally found to show poorer outcomes than parents of children without developmental disabilities concerning stress, well-being, and quality of life (Dabrowska & Pisula, 2010; Hamlyn-Wright, Draghi-Lorenz, & Ellis, 2007; Hedov, Anneren, & Wikblad, 2002; Marchal, Maurice-Stam, Hatzmann, Van Trotsenburg, & Grootenhuys, 2013; Roach, Orsmond, & Barratt, 1999). Compared with parents of children with other conditions associated with intellectual disability, however, the negative consequences seem relatively modest (Esbensen & Seltzer, 2011). Also, many parents reported to experience personal growth thanks to raising a child with DS (King & Patterson, 2000; Skotko, Levine, & Goldstein, 2011). Furthermore, most families of children with DS appear to show essentially normal family functioning (Cunningham, 1996; Povee, Roberts, Bourke, & Leonard, 2012), while divorce rates may be even lower than in the general population (Cunningham, 1996; Urbano & Hodapp, 2007). So, raising a child with DS appears to be a challenge, but one that many parents experience as worthwhile and that most families can adapt to.

Successful adaptation to the challenges of raising a child with DS depends on the resources of the family and one of these resources is support from health care professionals (Davis &

Gavidia-Payne, 2009). To provide targeted support, health care professionals should be aware of the type of practical and emotional problems that parents of children with DS are likely to encounter. For this purpose, studies of specific outcomes that relate to everyday life are needed, yet these studies are far less abundant than studies of parental perceptions and emotions (Hodapp, 2007). For example, we could only identify one study that systematically inquired specific everyday problems reported by parents of children with DS (Hedov, et al., 2002). This study, alongside studies of health related quality of life, suggests that the specific problems that parents struggle with focus on worries about the future of their child, time demands, sleep, vitality, social support and mental health (Bourke, et al., 2008; Hedov, Anneren, & Wikblad, 2000; Marchal, et al., 2013). This type of studies is essential if we want to understand the extent of consequences that parents experience in daily life as well as their need for practical assistance.

These previous studies of parental everyday problems and health related quality of life are limited in that they all concerned parents of children at early school age, or parents of children with DS in wide age ranges. Yet, the experience of parents may change substantially with age of the child (Cuskelly, Hauser-Cram, & Van Riper, 2008); several studies suggest that parents experience an increased burden as the child grows older (Hauser-Cram, et al., 2001; Hodapp, Ricci, Ly, & Fidler, 2003; Lanfranchi & Vianello, 2012). It is important, therefore, that studies of parental functioning take the age of the child into account, preferably employing a longitudinal design, or at least describing parental functioning within a limited age range of the children. Furthermore, in previous studies, mothers are overrepresented, even though the experiences of mothers and fathers raising a child with DS are not likely to be identical due to e.g. culturally defined expectations of family roles. Although recent studies found few differences between mothers and fathers of children with DS concerning, for instance, stress (Dabrowska & Pisula, 2010; Lanfranchi & Vianello, 2012), mothers and fathers do appear to differ in the type of stressors they experience. For mothers these concern fatigue, vitality, care responsibilities and social support, while fathers appear to experience more problems concerning attachment to their child, acceptability of their child to others, and financial consequences (Cuskelly, et al., 2008; Hedov, et al., 2000, 2002; Hodapp, 2007).

Beside knowledge of frequent problems at a group level, psychosocial screening or monitoring of individual families in clinical practice can ensure early detection and timely referral to targeted support (Kemper & Kelleher, 1996). Although psychosocial screening and monitoring have been successfully implemented in diverse medical settings (Haverman, et al., 2014; Valderas, et al., 2008), we could not identify reports of this in the care for families of children with DS. This is surprising given the potential psychosocial consequences of raising a child with DS. More knowledge about the outcomes of such screeners can provide practical and theoretical considerations for the use of such instruments in clinical practice.

In the current study we focused on parents of young adolescents (YAs) with DS and included

the perspectives of both mothers and fathers. We used a psychosocial screening questionnaire concerning distress and everyday problems that is frequently applied in Dutch clinical practice. We aimed to determine whether clinical distress and everyday problems were more frequent in mothers and fathers of YAs (11 to 13 year olds) with DS than in control parents of age-matched children without chronic disorders. Mothers and fathers were analysed separately to account for possible gender differences. Furthermore, we aimed to determine whether clinical distress and everyday problems differed between mothers and fathers in parent couples of YAs with DS.

## Methods

### Procedure and Respondents

Parents of YAs with DS were recruited among parents of children who had participated in a trial and a follow-up study concerning the effect of early thyroxine treatment in children with DS (Marchal et al., 2014). Participants for that trial were recruited among Dutch neonates with DS born between June 1999 and August 2001; exclusion criteria were an abnormal congenital hypothyroidism screening, premature birth, a 5-minute Apgar score below normal, or insufficient parental command of the Dutch language. During the follow up of this trial, between 2010 and 2012, parents had given permission to be invited for future studies. For the current study, parents received an email between December 2012 and March 2013 containing a description of the purpose of the study, a link to the study website, and unique login codes, one for each parent. After logging in, the parent could only start completing the questionnaire after giving informed consent. The study was performed with permission and in accordance with the regulations of the local Medical Ethical Committee.

Parents of age-matched YAs without DS were selected from norm data of the DT-P (van Oers et al., 2017). These parents were approached between November and December 2014 through the market research agency Taylor Nelson Sofres Netherlands Institute for Public Opinion (TNS-NIPO), which maintains a panel of around 200,000 potential respondents. To collect norm data for different questionnaires, a sample of parents of children aged 0-18 years was drawn from this panel that was representative of the Dutch population on key demographic characteristics. A total of 1423 parents responded, one parent per family. From this group, we selected parents who had a child aged  $\geq 11$  and  $< 14$  years, and who had no children with chronic conditions, as reported by parents. This resulted in a control group for the present study of 116 parents (64 mothers, 52 fathers) of YAs, aged 11-13 years, who had no chronic conditions.

### Measures

Responding parents of YAs with DS completed an online questionnaire concerning their socio-demographic characteristics: gender, country of birth (the Netherlands or other), educational

level (highest level completed, categorised afterwards as: low: primary, lower vocational, or lower or middle general secondary education; intermediate: middle vocational, higher secondary, or pre-university education; high: higher vocational education, or university), employment status (paid employment: yes or no), marital status, and number of children living at home. Parental age, and age and gender of their child with DS were known from the previous trial data.

The Distress Thermometer for Parents (DT-P) was used to assess overall distress and everyday problems (Haverman, et al., 2013). The DT-P is an adaptation of the Distress Thermometer, a screening tool used in standard adult oncology practice (Roth, et al., 1998). The adaptation consisted mainly of changes in problem items, i.e. deletion of several problem items concerning health and religion, and addition of problem items concerning cognitive functioning and parenting (Haverman, et al., 2013). The DT-P is a well-validated, brief screening instrument that is frequently used in clinical practice in the Netherlands. The DT-P serves as a screener to identify distress and everyday problems in parents of children with chronic conditions. The DT-P consists of a thermometer and a problem list. The thermometer asks parents to rate their overall distress on a scale from 0 (no distress) to 10 (extreme distress), with a score of 4 or higher indicating clinically elevated distress. The problem list inquires the occurrence over the past week (yes = 1, no = 0) of 34 everyday problems across six problem domains (practical, social, emotional, physical, cognitive, and parenting). Finally, four additional questions inquire perceived support from surroundings, perceived lack of understanding from people concerning their situation, parental chronic illness, and the wish to talk to a professional about their situation (yes, maybe, or no). From the problem items a problem domain score for each domain (total number of reported problems in the domain) and a total problem score (total number of reported problems) can be calculated. The internal consistency of the DT-P is acceptable with Cronbach's alphas ranging from .52 to .89 (van Oers, Schepers, Grootenhuis, & Haverman, 2017). In clinical practice the results of the DT-P as a screener are not only interpreted by the thermometer score, but also by the answers on the individual items. Therefore, outcomes of the DT-P were not only analysed by clinically elevated distress, total problem score, and problem domain scores, but also by problem items. Table 2 shows the everyday problem items as they are literally presented in the DT-P, except for the item concerning parental chronic illness which is listed as a socio-demographic characteristic in table 1.

### **Statistical Analysis**

First, preparatory analyses were performed: families of YAs with DS with at least one responding parent were compared with non-responding families concerning their child's gender (chi square test) and concerning the ages of the child and both parents (unpaired t tests). Next, socio-demographic characteristics of mothers and fathers of YAs with DS were compared with those of control mothers and fathers by unpaired t tests (age of parent, age of child with DS) and chi square/Fisher's Exact tests (remaining socio-demographic characteristics, see Measures).

**Table 1. Socio-demographic characteristics of mothers and fathers of young adolescents with Down syndrome compared with control mothers and fathers**

	Mothers			Fathers		
	DS N=76	Control N=64	p	DS N=44	Control N=52	p
<b>Parents</b>						
Age in years, M (SD)	45.9 (4.1)	42.2 (4.2)	<.001	47.8 (5.4)	46.7 (4.9)	.292
Born in the Netherlands, %	93.4	100	.063 <sup>a</sup>	97.7	100 <sup>c</sup>	.463 <sup>a</sup>
Educational level, % <sup>b</sup>			.180			.259
High	39.5	25.0		45.5	32.0 <sup>d</sup>	
Intermediate	40.8	53.1		43.2	46.0 <sup>d</sup>	
Low	19.7	21.9		11.4	22.0 <sup>d</sup>	
Paid employment, %	76.4 <sup>e</sup>	76.6	.981	90.5 <sup>d</sup>	94.2	.696 <sup>a</sup>
Marital status, %			.118			1.000 <sup>a</sup>
Married/living together	93.4	82.8		97.7	98.1	
Single/separated	6.6	15.6		2.3	1.9	
Undisclosed	0	1.6		0	0	
Children living at home, %			.211			.165
1	10.5	20.3		11.4	15.4	
2	53.9	53.1		54.5	67.3	
≥ 3	35.5	26.6		34.1	17.3	
Chronic illness, %	9.2	21.9	.037	11.4	25.0	.088
<b>Child</b>						
Age in years, M (SD)	12.6 (0.7)	12.4 (0.9)	.141	12.7 (0.6)	12.3 (0.9)	.011
Female gender, %	46.1	50.0	.641	45.5	51.9	.528

Notes. The group of parents of young adolescents with DS consisted of 120 parents of 86 children (34 parent couples). Characteristics were compared by unpaired t tests for continuous variables and by chi square or Fisher's Exact Test (indicated with <sup>a</sup>) for categorical variables.

<sup>b</sup> Highest educational level completed. High: higher vocational education, university; Intermediate: middle vocational education, higher secondary education, pre-university education; Low: Primary education, lower vocational education, lower or middle general secondary education.

<sup>c</sup> Score of 1 parent missing.

<sup>d</sup> Score of 2 parents missing.

<sup>e</sup> Score of 4 parents missing.

Subsequently, to determine whether it would be necessary to include socio-demographic characteristics in the analyses of clinical distress, we tested which socio-demographics related to clinical distress (in mothers and fathers, and in DS- and control parents separately). Variables on which parents with clinical distress scores differed from parents without clinical distress scores (at a significance level of 0.20, based on unpaired t tests [age of parent] or chi square/Fisher's Exact tests [remaining socio-demographic characteristics and parental chronic illness, see Measures]), were included in a logistic regression of clinical distress. For mothers and fathers of YAs with DS and for control fathers, none of the socio-demographic characteristics contributed significantly to the regression model of clinical distress (data not shown). For



control mothers, however, parental chronic illness (OR 9.24, 95% CI 1.72 to 49.82) related to clinical distress. After this, we compared socio-demographics of parents of YAs with DS and control parents (mothers and fathers separately), which revealed that parental chronic illness was more frequent among control mothers. Given the latter difference and the relation of parental chronic illness with clinical distress in control mothers, it was added as covariate in the comparison of the DT-P outcomes between parents of YAs with DS and control parents..

Next, we analysed DT-P outcomes by: a) clinically elevated distress, b) total problem score, c) problem domain scores, and d) problem items. This was done first in parents of YAs with DS versus control parents, separately analysing mothers and fathers. Logistic regression analyses, with parental chronic illness as covariate, were used to test differences between parents of YAs with DS and their controls concerning: a) clinically elevated distress and d) problem items. Analysis of variance (ANOVA), with parental chronic illness as covariate, was performed to test differences between parents of YAs with DS and their controls on: b) total problem score and c) problem domain scores.

Finally, we compared DT-P outcomes in mothers and fathers from parent couples of YAs with DS; McNemar tests were used to test differences concerning a) clinically elevated distress and d) problem items. Paired t tests were used to test differences between mothers and fathers concerning b) total problem score and c) problem domain scores.

In this study we aimed to explore the everyday problems of mothers and fathers of YAs with DS. Therefore, we preferred avoiding type 2 errors over avoiding type 1 errors. Consequently, we did not correct for multiple testing, using a significance level of  $p < .05$ . Given that we compared 45 different outcomes per group, around two to three false positive findings are expected at this significance level per group comparison. All data in this study were analysed using SPSS version 24.

## Results

### Preparatory analyses

Of the invited parents of 123 YAs with DS, parents of 86 YAs with DS responded (response rate 70%). This concerned 120 parents: 76 mothers and 44 fathers, including 34 parent couples. Their children with DS were aged 11.5 to 13.7 years and 52.3% of the children were boys. Families of YAs with DS with at least one responding parent did not differ significantly from non-responding families regarding the compared socio-demographics (data not shown). Table 1 shows socio-demographics of parents of YAs with DS and control parents; mothers of YAs with DS were significantly older, but reported fewer chronic illnesses than control mothers. Furthermore, fathers of YAs with DS had significantly older children than control fathers. These variables (parental age, parental chronic illness, and age of the child with DS) were checked

**Table 2. Outcomes of the Distress Thermometer for Parents of mothers and fathers of young adolescents with Down syndrome compared with those of control mothers and fathers**

	Mothers				Fathers			
	DS N=76	Control N=64	p	OR/ES 95% CI	DS N=44	Control N=52	p	OR/ES 95% CI
<b>Thermometer score</b>								
Clinical, %	46.1	48.4	.741	1.13	36.4	44.2	.715	0.85
<b>Total problem score, M (SD)</b>	7.5 (6.7)	6.3 (6.4)	.142	0.19	6.3 (5.9)	3.5 (4.8)	.005	0.58
<b>Practical problems, M (SD)</b>	1.1 (1.3)	1.3 (1.5)	.689	-0.13	1.0 (1.3)	0.7 (1.2)	.042	0.25
Housing, %	3.9	12.5	.130	0.34	2.3	3.8	.943	0.91
Work/study, %	17.1	35.9	<b>.017</b>	<b>0.38</b>	31.8	19.2	.111	2.21
Finances/insurance, %	14.5	18.8	.530	0.75	18.2	9.6	.148	2.53
Housekeeping, %	28.9	20.3	.173	1.76	11.4	11.5	.675	1.33
Transport, %	7.9	9.4	.892	0.92	2.3	5.8	.552	0.49
Child care/child supervision, %	9.2	6.2	.477	1.60	9.1	5.8	.450	1.85
Leisure activities/relaxing, %	32.9	26.6	.373	1.41	29.5	9.6	<b>.013</b>	4.43
<b>Social problems, M (SD)</b>	0.5 (0.8)	0.5 (0.9)	.509	0.00	0.5 (0.8)	0.3 (0.6)	<b>.032<sup>a</sup></b>	0.33
Dealing with (ex)partner, %	25.0	12.5	<b>.038</b>	<b>2.73</b>	18.2	13.5	.348	1.74
Dealing with family, %	13.2	10.9	.550	1.38	9.1	3.8	.223	3.10
Dealing with friends, %	2.6	6.2	.404	0.47	9.1	0.0	.997	-
Interacting with your child(ren), %	10.5	17.2	.366	0.63	13.6	7.7	.258	2.23
<b>Emotional problems, M (SD)</b>	2.1 (2.3)	1.8 (2.3)	.304	0.13	1.8 (2.3)	1.0 (1.6)	<b>.041<sup>a</sup></b>	0.50
Keeping emotions under control, %	31.6	26.6	.302	1.50	27.3	9.6	<b>.049</b>	3.17
Self-confidence, %	27.6	23.4	.464	1.34	15.9	9.6	.447	1.62
Fears, %	13.2	10.9	.550	1.38	11.4	7.7	.401	1.85
Depression, %	27.6	37.5	.250	0.65	38.6	23.1	<b>.040</b>	2.74
Feeling tense or nervous, %	36.8	31.2	.336	1.43	31.8	26.9	.684	1.20
Loneliness, %	15.8	12.5	.347	1.63	13.6	1.9	.060	8.16
Feelings of guilt, %	14.5	10.9	.352	1.65	4.5	5.8	.795	0.78
Use of substances (e.g. alcohol, drugs and/or medication), %	2.6	3.1	.940	0.92	9.1	3.8	.151	4.03
Intrusive/recurrent thoughts about a specific event, %	35.5	23.4	.068	2.07	31.8	15.4	<b>.045</b>	2.83



for their relation with a clinical thermometer score. This revealed that of these variables, only parental chronic illness showed a significant relation with a clinical thermometer score, both in the combined group of mothers  $\chi^2(1, N = 140) = 11.33, p < .001$  and in the combined group of fathers,  $\chi^2(1, N = 96) = 6.23, p = .013$ . This indicated that parents with a chronic illness (which were more frequent in the control group) more often reported clinical distress than parents without chronic illness. Therefore, parental chronic illness was included as covariate in the comparisons of DT-P outcomes in parents of YAs with DS versus parents of YAs without DS.

### DT-P outcomes

Table 2 presents DT-P outcomes in mothers and fathers of YAs with DS as compared with their controls. Clinically elevated distress was not more frequent in parents of YAs with DS than in control parents, in both mothers and fathers. The total problem score was not higher in mothers of YAs with DS versus control mothers, but was higher in fathers of YAs with DS versus control fathers.

Concerning the problem domain scores, mothers of YAs with DS only reported a higher number of problems on the cognitive domain, specifically concerning concentration and memory. The analyses of the everyday problem items and additional items revealed that mothers of YAs with DS reported more frequent problems than control mothers concerning dealing with their (ex)partner (social domain), independence of their child (parenting domain), receiving enough support from people around them, and people reacting with a lack of understanding to their situation (both additional items). Concerning work/study (practical domain), mothers of YAs with DS reported fewer problems than control mothers.

Concerning the problem domain scores, fathers of YAs with DS scored higher than control fathers in all problem domains except the parenting domain. Within the practical domain they reported more problems with leisure activities/relaxing. Even though fathers reported a higher number of social problems they did not differ significantly from their controls on any of the items in the social domain. Within the emotional domain, fathers of YAs with DS reported more problems with keeping emotions under control, depression, and intrusive or recurrent thoughts about a specific event than control fathers. Within the physical domain weight and sexuality were more frequently a problem in fathers of YAs with DS than in control fathers. Like mothers, fathers reported more cognitive problems than their controls, specifically concerning concentration and memory. Furthermore, even though the number of parenting problems was not higher in fathers of YAs with DS, they did report more problems than their controls with the child's independence. Regarding the additional items, fathers of YAs with DS reported more often than control fathers to have problems with receiving enough support from people around them and more frequently indicated to (maybe) want to talk to a professional about their situation.

The comparison of mothers with fathers from 34 parent couples of YAs with DS revealed no significant differences concerning clinical distress, total number of problems, problem domain scores, or problem items (supplemental table 1).

## Discussion

This study compared outcomes of a psychosocial screening questionnaire for distress and everyday problems in 120 parents of YAs (11 to 13-year-olds) with Down syndrome versus control parents of age-matched YAs without chronic conditions. Clinical distress was not more frequently found in parents of YAs with DS than in control parents. Mothers did not report a higher total number of everyday problems than control mothers and only reported more problems than their controls in one domain (cognitive problems) as well as on some items concerning partner relation and social support. Meanwhile they reported less problems concerning work or study. Fathers, however, reported more problems than their controls, overall and in all domains except parenting. Furthermore, fathers more frequently wished to talk to a professional about their situation than control fathers. Within parent couples, mothers and fathers did not show statistically significant different DT-P outcomes.

A first striking finding is that in terms of distress, as indicated by the frequency of clinical distress, parents of YAs with DS did not differ from control parents. Moreover, among mothers of YAs with DS, everyday problems were not more frequent than among their controls. In essence, mothers hardly differed from their controls on this psychosocial screener, which is an encouraging finding. Our results are another indication that many parents are resilient and that raising a child with DS is not an inevitable tragedy (Povee, et al., 2012; Van Riper, 2007).

Nevertheless, mothers reported some distinct problems more frequently than their controls. Problems that were reported by mothers as well as by fathers may represent relevant everyday problems in these families. First, mothers and fathers both reported more problems than their controls in the cognitive domain. These problems may reflect objectively measurable cognitive functioning (Bender, et al., 2008), but may also reflect mental health issues such as anxiety or depression (Elixhauser, Leidy, Meador, Means, & Willian, 1999; Lovell, Elliot, Liu, & Wetherell, 2014; Middleton, Denney, Lynch, & Parmenter, 2006) which would be in line with the previously found poorer mental health scores in health related quality of life studies in parents of children with DS (Bourke, et al., 2008; Hedov, et al., 2000). In clinical practice, when an individual reports cognitive problems, further exploration of possible causes is indicated. Second, mothers and fathers both reported problems with (social) support from their environment, which is an important factor for parental and family functioning (Cantwell, Muldoon, & Gallagher, 2014; Gavidia-Payne, Denny, Davis, Francis, & Jackson, 2015; Marchal, et al., 2013; Van Oers, et al., 2014). In addition, mothers (but not fathers) reported more frequent problems in dealing with

their (ex)partner, which is another important factor for well-being of the parent and of the family (Kersh et al. 2006, Marchal et al. 2013, Norlin and Broberg 2013). Previous literature provided few indications that partner relation problems are a specific issue for parents of children with DS (Van Riper et al. 1992, Povee et al. 2012). Yet, when they are reported by families, problems with social support and the partner relation are important to address to ensure prospective well-being. Another problem that both mothers and fathers reported more frequently, concerned their child's independence. This is not surprising given the developmental disability in children with DS and may relate to the worries of parents about the future of their child, which was identified as an important theme in a previous study of parental everyday problems (Hedov, et al., 2002).

Interestingly, both mothers and fathers did not report more problems with sleeping than their respective controls, whereas this was previously found in parents of younger children with DS (Hedov, et al., 2002); this may reflect a decline in sleep problems in their children with DS by the adolescent age (Breslin, Edgin, Bootzin, Goodwin, & Nadel, 2011). It is also of interest that mothers of YAs with DS reported less problems than their controls with work or study than control mothers. This may reflect a tendency to choose for a less demanding career, to be able to combine work with the care for their child, as similarly found in a Swedish study (Hedov et al. 2002). These career choices of mothers of children with DS deserve further exploration.

Although mothers of YAs with DS did not appear to differ much from control mothers, fathers showed less favourable outcomes than control fathers concerning everyday problems. Almost a third of fathers of YAs with DS indicated to (maybe) want to talk to a professional about their situation, underscoring that professional help may be needed for these fathers. Fathers reported more frequent problems than their controls across a range of domains and items; beside the problems concerning cognition, child's independence, and support from surroundings that were discussed above, fathers reported a higher number of social problems. Since none of the problem items in this domain revealed a significant difference from controls, this appears to represent an overall lack of time and flexibility to invest in social activities, which may relate to the care demands (Wayne & Krishnagiri, 2005). Similarly, fathers reported more problems with leisure than control fathers. Furthermore, fathers reported more problems in the physical domain, specifically concerning weight and sexuality. Problems with sexuality are likely related to the partner relation, which mothers more frequently reported to be a problem (Young, Denny, Luquis, & Young, 1998). Yet, problems with weight and sexuality are too complex to be explained by a single factor and can both be placed in a biopsychosocial model (Althof, et al., 2005; Skelton, DeMattia, Miller, & Olivier, 2006). As such, they may also be linked to emotional problems that fathers currently reported (Kivimaki, et al., 2006; Luppino, et al., 2010; Nicolosi, Moreira, Villa, & Glasser, 2004; Torres & Nowson, 2007). Whatever the precise underlying mechanisms are, these physical problems likely represent a diminished feeling of vitality, as was found in previous studies concerning health related quality of life (Bourke, et al., 2008; Hedov, et al., 2000; Marchal, et al., 2013).

In sum, our findings point in an unexpected direction. Based on previous literature, the finding that mothers hardly differ from control mothers, while fathers show unfavourable outcomes compared with control fathers, is surprising. The content of the problems that fathers reported more frequently than their controls, was less surprising; apart from the cognitive problems, they were consistent with issues that were previously found in parents of younger children with DS (i.e. worries about the child's future, time demands, vitality, social support, and mental health).

Notwithstanding the relatively favourable outcomes in mothers of YAs with DS and the relatively unfavourable outcomes in fathers, there was no statistically significant difference between mothers and their spouses. We only found a tendency towards mothers reporting more housekeeping and fatigue problems, in a direction as expected based on previous findings that mothers of children with DS experience more problems with fatigue and care responsibilities than fathers (Cuskelly et al. 2008, Hodapp 2007, Hedov et al. 2002, Hedov et al. 2000). In the recently published norm data of the DT-P, fathers had less distress and fewer problems than mothers (van Oers, et al., 2017). Therefore, the finding that fathers of YAs with DS did not show less distress and fewer problems than their spouses may be seen as another indication of relatively unfavorable outcomes in fathers. To determine whether the (absence of) differences between mothers and fathers are similar to what is found in mothers and fathers in the general population, we would have to compare mothers and fathers from parent couples in the control group also. Unfortunately, we were unable to do so, since only one parent per family responded in the control group.

The aim of implementing the DT-P or similar screeners in clinical practice, is early identification of psychosocial issues. In about half of parents of YAs with DS no further exploration of their situation was indicated, since they had neither a clinical distress score nor a wish to talk to a professional (data not shown). Even though psychosocial screening results among mothers were very similar to those of controls, timely detection of everyday problems or psychosocial issues and allocation of support to those who need it most, may still be relevant. Even in the general population most mental health problems are left untreated, while timely intervention is often cost-effective (Demyttenaere et al., 2004). As such, psychosocial screening instruments can play an important role in standard care for families of children with DS. The current study was in an experimental setting, future studies should report on what implementation of such screening in clinical practice yields, with particular focus on prospective preventive effects.

The current study is one of few studies that systematically inquired everyday problems in parents of YAs with DS and the first to do so using a commonly applied psychosocial screening tool. Furthermore, we were able to incorporate the experiences of both mothers and fathers, which shed light on potential differences in their experiences. Our study does, however, have some limitations. First, for the purpose of the trial from which parents were recruited, children with DS who were born prematurely, who had abnormal thyroid screening results, who had an Apgar score below normal, and whose parents had insufficient command of the Dutch

language, were excluded from participation. Therefore, our sample represents a relatively 'healthy' subgroup of YAs with DS, whose parents may experience less everyday problems than parents of YAs with DS who would have more comorbid health problems. Furthermore, it should be kept in mind that there is a chance of false positive findings in the current study, since we chose to not correct for multiple testing. In mothers, we found that eight out of 45 comparisons yielded significant differences, in fathers this was 17 comparisons of 45. Given the expected two or three 'random' significant differences, most reported results are likely not by chance alone. Also, the modest number of respondents limited the statistical power and the possibilities of statistical methods for this study. Finally, given the setting of this study, our results represent the specific Dutch situation with its specific culture and regulations concerning e.g. health insurance and education.

In conclusion, mothers of YAs showed few differences from control mothers on a screening instrument concerning distress and everyday problems. Fathers, however, reported substantially more everyday problems than their controls and more frequently wished to talk to a professional about their situation. This implies that fathers' psychosocial functioning should receive attention in the care for families of YAs with DS. Our findings further imply that to detect psychosocial problems at an early stage, inquiring parents, and fathers in particular, about their experienced distress may not suffice. Rather, discussing specific problems and the wish for referral seems required. This is suggested by the discrepancy between the 'normal' levels of clinical distress in fathers, whilst they reported more everyday problems than control fathers and more frequently wished to talk to a professional about their situation. This discussion can be facilitated in clinical practice by screening instruments such as the DT-P.

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## Supplemental table 1

## Distress Thermometer score, problem domain scores and problem item scores in parent couples of young adolescents with DS: mothers compared with fathers

	Mothers (N=34)	Fathers (N=34)	p
<b>Thermometer score</b>			
Clinical (%)	47.1	41.2	.791
<b>Total problem score, M (SD)</b>	6.9 (6.7)	6.2 (5.9)	.548
<b>Practical problems, M (SD)</b>	1.3 (1.3)	1.0 (1.3)	.412
Housing, %	5.9	2.9	1.000
Work/study, %	23.5	32.4	.607
Finances/insurance, %	17.6	23.5	.727
Housekeeping, %	29.4	11.8	.070
Transport, %	8.8	2.9	.625
Child care/child supervision, %	11.8	5.9	.687
Leisure activities/relaxing, %	29.4	23.5	.774
<b>Social problems, M (SD)</b>	0.6 (0.9)	0.4 (0.8)	.228
Dealing with (ex)partner, %	26.5	17.6	.508
Dealing with family, %	17.6	8.8	.508
Dealing with friends, %	5.9	2.9	1.000
Interacting with your child(ren), %	17.6	11.8	.625
<b>Emotional problems, M (SD)</b>	1.8 (2.3)	1.9 (2.3)	.910
Keeping emotions under control, %	32.4	29.4	1.000
Self-confidence, %	29.4	20.6	.581
Fears, %	11.8	11.8	1.000
Depression, %	26.5	38.2	.424
Feeling tense or nervous, %	32.4	35.3	1.000
Loneliness, %	11.8	11.8	1.000
Feelings of guilt, %	8.8	2.9	.625
Use of substances (e.g. alcohol, drugs and/or medication) , %	0.0	5.9	-
Intrusive/recurrent thoughts about a specific event, %	29.4	32.4	1.000
<b>Physical problems, M (SD)</b>	2.0 (1.9)	1.6 (1.6)	.255
Eating, %	8.8	2.9	.625
Weight, %	23.5	32.4	.508
Sleep, %	38.2	32.4	.791
Fatigue, %	58.8	35.3	.096
Out of shape/condition, %	23.5	14.7	.453
Pain, %	35.3	26.5	.581
Sexuality, %	14.7	20.6	.625
<b>Cognitive problems, M (SD)</b>	0.5 (0.8)	0.4 (0.7)	.872
Concentration, %	23.5	26.5	1.000
Memory, %	23.5	17.6	.754
<b>Parenting problems, M (SD) <sup>c</sup></b>	0.7 (1.2)	0.7 (0.9)	.751
Dealing with your child, %	11.8	2.9	.375
Dealing with the feelings of your child, %	11.8	14.7	1.000
Talking about the disease/consequences with your child, % <sup>d</sup>	8.8	11.8	1.000
Independence of your child, %	32.4	41.2	.508
Following advice about treatment/giving medication, %	2.9	2.9	1.000
<b>Additional questions, n (%)</b>			
Do you feel you receive enough support from people around you?, %	73.5	79.4	.727
Do people often react to your situation with a lack of understanding, %	23.5	20.6	1.000
Would you like to talk to a professional about situation - Yes/Maybe, %	35.3	20.6	.180

Notes. Total problem score and problem domain scores were analysed with paired t tests. The presence of a clinical thermometer score and of reported problems (everyday problem items) were analysed with McNemar tests.