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Parenting Children With a Cleft Lip With or Without Palate or a Visible Infantile Hemangioma: A Cross-Sectional Study of Distress and Parenting Stress

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Abstract

Objective: Parents of children with a medical condition and a visible difference can experience challenging situations. We evaluated distress and parenting stress in parents of children with a cleft lip with or without cleft palate (CL±P) or a visible infantile hemangioma (IH).

Setting: This cross-sectional study took place in an academic medical hospital in Rotterdam, the Netherlands.

Participants: Three-hundred nine parents (mean age = 40.30, 56.00% mothers) of children with CL±P and 91 parents (mean age = 36.40, 58.24% mothers) of children with IH.

Main Outcome Measures: The Dutch version of the Parenting Stress Index – Short Form and the subscales Anxiety, Depression, and Hostility of the Symptom Checklist – 90.

Results: One sample *t* tests and mixed linear modeling were used. On average, parents of children with CL±P and of children with IH showed significantly lower parenting stress compared to normative data. Anxiety was significantly lower in parents of children with CL±P than that in the norm group. Visibility of the condition was not related to distress or parenting stress. Child behavioral problems were positively related to parenting stress, depression, and hostility.

Conclusions: Parents of children with CL±P and IH report less distress and parenting stress compared to the norm. On average, these parents seem well adjusted. A practical implication is to monitor parents of children with behavioral problems.

Keywords

psychological stress, parents, cleft lip, infantile hemangioma

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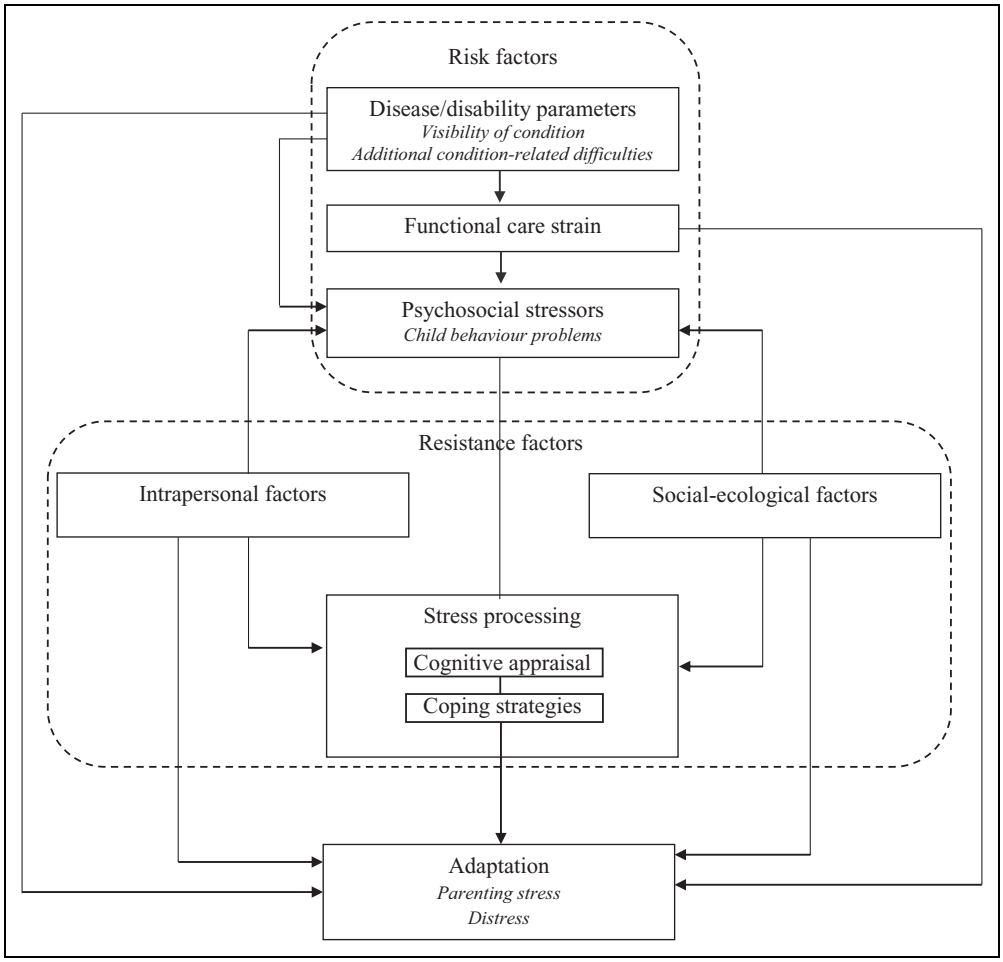

Figure 1. Model posited by Wallander et al. (1989). Constructs measured in the current study in italics.

Introduction

Parenting children with a medical condition, such as a cleft lip with or without cleft palate (CL ± P) or an infantile hemangioma (IH), can present significant challenges. Apart from having to face medical treatment and possible hospitalizations, the visibility of these conditions means that these parents may have to cope with negative social feedback related to the child's appearance (Klein et al., 2006; Klein et al., 2010). Hence, raising a child with a visible difference due to a medical condition can have an impact on parental psychological adaptation.

In an attempt to explain how parents adapt to having a child with a chronic condition and/or illness, Wallander et al. (1989) posited a model of risk and resistance factors that have a direct impact on parental adaptation. An adaptation of this model is shown in Figure 1 along with the constructs tested in the current study. Furthermore, risk factors have an indirect influence on parental adaptation through stress processing (ie, cognitive appraisal and coping strategies). The current study focuses on the direct impact of risk factors on parental adaptation, conceptualized as distress and parenting stress, in parents of children with CL ± P or IH. Both conditions are present from birth or very shortly afterward and may involve frequent hospital visits. In addition, these conditions become less visible over time, either due to surgery (CL ± P) or due to the natural course of the condition itself (IH). Both conditions may also leave permanent marks on the face. In CL ± P, a scar may be visible (Goodacre & Swan, 2012). Infantile hemangiomas regress over time, with the majority of the regression occurring before 4 years of age (Darrow et al., 2015) and 50% to 70% of IHs eventually resolve. However, residual skin changes such as scars or redundant skin can be present (Darrow et al., 2015).

Parenting stress has been defined as "a set of processes that lead to aversive psychological and physiological reactions arising from attempts to adapt to the demands of parenthood" (Deater-Deckard & Scarr, 2004). Parenting distress has been defined as "the extent to which the parent perceives stress in his or her role as a parent" (Abidin & Abidin, 1990). Although specific measures exist for measuring parenting stress (eg, the Parenting Stress Index [PSI]; Abidin, 1983), parenting distress is most often conceptualized as parental depression or anxiety (Deater-Deckard & Panneton, 2017). When looking at parents of children with a chronic illness in general, recent meta-analyses (Pinquart, 2018b, 2018a) showed that parents of

children with a chronic physical illness experience more parenting stress, anxiety, and depression compared to parents of healthy children or normative data. For parents of children with craniofacial conditions or CL ± P, no significant differences were found on parenting stress, parental anxiety, and parental depression compared to normative data or a control group (Pinquart, 2018b, 2018a). Parents of children with IH were not included in these meta-analyses. Although visibility was not investigated across diagnoses, moderate elevations of parental anxiety and depression were reported for skin diseases, which are often visible. A recent review and meta-analysis has shown that adolescents with a visible difference experience more anxiety than unaffected peers (van Dalen et al., 2020). Rumsey et al. (2004) showed that 47% of adults with a visible difference have subclinical or clinical symptoms of anxiety. However, less is known about the impact of the visible difference on parents. Qualitative studies showed parents have to manage other people's questions and reactions to their child's altered appearance (Barke et al., 2016). Hoornweg et al. (2009) reported that parents' quality of life is not associated with the visibility of the child's hemangioma. However, to date, no literature exists that quantitatively examines the impact of the visible difference on parents' stress and distress.

Literature on distress and parenting stress in CL ± P, reflecting parental adaptation, is scarce. In a study by Hasanzadeh et al. (2014), 55 mothers of children aged 8 to 18 years with CL ± P participated. Of these mothers, 38.2% experienced psychological distress and 23.6% showed signs of serious psychological problems, such as depression and anxiety. A recent study by Stock et al. (2020) among 1163 parents of children with cleft lip and/or palate (CL/P) showed that mothers of newborn children with CL/P score higher on anxiety and depression when compared to normative data. Fathers scored lower on anxiety but higher on depression when compared to normative data. Nevertheless, small effect sizes were reported for all findings in this study and visible clefts were not reported separately from the nonvisible clefts. Another study found no significant differences on parenting stress between parents of children with CL ± P and controls (Collett et al., 2012). There is also literature showing that parents of children with CL ± P experience low levels of distress and a high degree of positive adjustment (Baker et al., 2009). Literature on IH is less common. Cazeau et al. (2017) reported that 70% of parents are psychologically impacted by the child's IH, for instance, through the gaze of others and experiencing anxiety, and that only 10% were offered psychological support.

This study quantitatively investigated distress and parenting stress in parents of children with CL ± P or IH. This is important, as previous literature has shown that parenting stress can lead to child behavioral problems (Neece et al., 2012). Furthermore, parents' distress and stress can be transferred to the child through emotional contagion (Hatfield et al., 1993). We sought to answer 2 research questions: (1) Do parents of children with CL ± P or IH report higher levels of distress and parenting stress than parents from the general population? (2) How are parent-perceived visibility and other parent-reported factors related to distress and parenting stress in parents of children with CL ± P or IH? We examined the roles of parent-perceived visibility, medical problems related to the condition, child behavioral problems, parent age, child age, and child gender in parental adjustment. As earlier research has shown that subjective visibility (ie, the visibility as experienced by the child or parents) of the condition is a much stronger predictor of adjustment in the person with a visible difference than "objective" visibility (ie, the visibility as judged by a physician; Moss, 2005), we focused only on subjective visibility as experienced by parents. Following others (Hasanzadeh et al., 2014; Cazeau et al., 2017; Stock et al., 2020), we hypothesized that parents of children with CL ± P or IH would experience more distress and parenting stress than other parents, as they face many different challenges. As there is minimal literature specifically assessing the influence of subjective visibility on distress and parenting stress, we did not have any prior hypotheses concerning research question 2.

Methods

Sample and Procedure

This study was part of a larger project assessing parental well-being and behavior of parents with children with CL ± P or IH. The study was conducted in accordance to the Declaration of Helsinki (World Medical Association, 2013).

Recruitment took place in the Centre of Paediatric Dermatology of the Department of Dermatology and the Department of Oral and Maxillofacial surgery of the Erasmus MC-Sophia Children's Hospital in Rotterdam, the Netherlands. Recruitment took place from 2008 until 2011, and data were later analyzed and reported in 2019 and 2020 by a PhD student. Inclusion criteria were (1) sufficient knowledge of the Dutch language by parents and (2) having a child with a visible CL ± P or IH, aged between 0 and 12 years. This as range was chosen as, in the Netherlands, children often make the transition to secondary education at age 12. For many parents and their children, this is seen as the end of childhood and the start of adolescence. Children with an isolated cleft palate or cleft alveolus were not eligible for this study, due to the nonvisible nature of these conditions. Each eligible family received an informed consent letter, 2 sets of questionnaires and a prepaid envelope. Parents were asked to fill out the questionnaires separately and independently from their partner. As an incentive, parents received a small gift for their child, worth approximately €1,-. Reminders were sent approximately 1 month later and again after 3 months.

Medical Care At The Erasmus MC-Sophia Children's Hospital, The Netherlands

Cleft care was provided at the Erasmus MC-Sophia Children's Hospital, the Netherlands, by multidisciplinary teams, with average cleft lip surgery at the age of 3 months and cleft palate closure at the average age of 12 months. The alveolar cleft is closed around 9 to 12 years of age. Access to speech therapy is

guaranteed through the treating hospital. Patients are followed up regularly by the multidisciplinary team until age 22.

Medical care for patients with an IH was provided at the Centre for Congenital Vascular Anomalies of the Erasmus MC. Treatment was administered (such as propranolol topical/systemic, prednisone, system/intralesional, surgery) to children with IH. All treated IHs were either potentially (life-)threatening or had functional risk, local discomfort, or cosmetic consequences. In case of treatment, follow-up is until the end of systemic treatment. If cosmetic surgery is a suspected possibility, the IHs or its residuals are reevaluated at age 3.

Instruments

Parent and child characteristics. Demographic and participant characteristics were obtained using questionnaires and included age and ethnicity of children and parents and parental educational level as a proxy measure of socioeconomic status. Parental education was divided into low, middle, and high, based on the International Standard Classification of Education (ISCED) guidelines (CBS, 2011). These guidelines provide an international classification for organizing education programs by education levels. It consists of 9 education levels (ISCED, 2011). Parents with primary or lower secondary education were coded as low. Parents with upper secondary, postsecondary nontertiary, or short-cycle tertiary education were classified as average. Examples of these programs include vocational certifications and associate's degrees. Parents with a bachelor's degree or higher were classified as high.

Parenting stress. The NOSI-K (de Brock et al., 1992) is the Dutch, shortened version of the PSI (Abidin, 1983). The NOSI-K is designed to measure parenting stress in parents of children aged 2 to 13 years; however, the NOSI-K has also been used in studies of infants younger than 2 years (eg, van der Pal et al., 2008; Meijssen et al., 2011). Participant results must be interpreted with caution, given the context of the younger age range in the IH group from the norm groups. The scale consists of 25 items rated on a 6-point scale, ranging from 1 (*totally disagree*) to 6 (*totally agree*). A total parenting stress score is obtained by summing the ratings across items, with high scores reflecting high levels of parenting stress. The normative data of the NOSI-K consist of 161 mothers and 84 fathers (de Brock et al., 1992). Scores can be classified into 7 categories, ranging from extremely low to extremely high.

The NOSI-K discriminates well between clinical and nonclinical samples (de Brock et al., 1992) and has acceptable internal consistency in other studies (van der Pal et al., 2008; van der Veek et al., 2009). In the present study, Cronbach $\alpha = .94$ for the CL ± P group and $\alpha = .95$ for the IH group.

Distress. Distress experienced by parents was operationalized by symptoms of depression, anxiety, and hostility as measured by the corresponding subscales of the Dutch translation of the Symptom Checklist – 90 (SCL-90; Arrindell & Ettema, 1986). Depression and anxiety capture internalizing problems, whereas hostility captures externalizing problems such as feelings of anger and aggression. Hostility was included as clinical practice learns that parents may experience resentment or angry feelings following their child's diagnosis (Nelson et al., 2012). The SCL-90 is a 90-item multidimensional questionnaire designed to screen for a broad range of psychological problems. The 16-item Depression (eg, feeling lonely), 10-item Anxiety (eg, suddenly scared for no reason), and 6-item Hostility (eg, having urges to beat, injure, or harm someone) subscales were used. They are rated on a 5-point Likert scale, ranging from 1 (*not at all*) to 5 (*very much*). Scale scores are calculated by summing the item scores, with higher scores reflecting higher levels of depressive feelings, anxiety, and feelings of hostility.

Adequate psychometric properties have been found in a sample of 2366 adults from the general population (Arrindell & Ettema, 1986). In the present study, Cronbach α for measures of parenting distress ranged between $\alpha = .76$ and $\alpha = .92$. The subscale hostility for IH ($\alpha = .76$) was the only subscale below $\alpha = .80$.

Child behavioral problems. The Child Behavior Checklist: Ages 1½ to 5 (CBCL 1½-5; Achenbach & Rescorla, 2000) and the Child Behavior Checklist: Ages 6 to 18 (CBCL 6-18; Achenbach & Rescorla, 2001) are widely used parent-reported measures of childhood behavioral problems and competencies. The CBCL 1½-5 consists of 100 items, the CBCL 6 to 18 contains 120 items. Both are scored on a 3-point scale, ranging from 0 (*not true*) to 2 (*very true or often true*). The Total Problems scale is obtained by summing the item scores, with high scores reflecting higher levels of behavioral problems. Because the 2 CBCL age versions differ in number in items, a 120-item equivalent of CBCL 1½-5 total scores was computed. As with the NOSI-K, some participants in the IH group were younger than the CBCL 1½-5 was developed to measure; however, total raw scores rather than norm comparisons were used in interpreting the CBCL results. In the present study, Cronbach α ranged between $\alpha = .93$ and $\alpha = .97$.

Visibility. To assess subjective visibility of the condition, parents were asked 2 questions, both rated on a 5-point Likert scale: (1) "To what extent do you think your child's condition is visible?" and (2) "To what extent do you feel that bystanders look at your child's visible difference?" (both ranging from *not at all* to *very much*). The 2 items were summed to derive a total visibility score. The items correlated significantly ($r = 0.59$, $P < .001$).

Additional condition-related difficulties. Additional questions were posed for potential medical or adjustment problems ("Does your child have feeding problems?," "Does your child have speech problems?," "Does your child have learning difficulties?," and "Does your child have psychological problems? [eg, Is your child aggressive? Is your child depressed?]"). All questions were rated on a 5-point Likert scale and summed to derive a total score.

Statistical Analysis

The means (Ms) and SDs of all variables were calculated. In preliminary analyses, the demographic variables of the CL ± P versus IH group were compared using χ^2 tests for categorical variables and independent samples *t* tests for continuous variables. The Levene's test was used to assess equal variances for both groups in the independent samples *t* tests. If this test is significant, SPSS automatically corrects the degrees of freedom.

For research question 1, one-sample *t* tests were used to compare scores on the NOSI-K and SCL-90 with the normative mean scores. Cohen *d* was calculated as a measure of effect size. Following Cohen (1992), $d = 0.20$ was considered a small effect, $d = 0.50$ medium, and $d = 0.80$ large.

Research question 2 was tested using linear mixed models to compare distress and stress measures (NOSI-K, SCL-90 anxiety, depression, and hostility) among the CL ± P and IH groups. This analysis accounts for clustering in the data (ie, mothers and fathers reporting on the same child). Separate analyses were conducted for each outcome measure of distress or stress. Parent and child age, parent and child gender, child behavior problems, visibility of the condition, parent education, and other difficulties related to the condition were used as covariates. To assess whether cleft type influenced distress and parenting stress, secondary analyses were run. Mixed linear models were computed as described previously; however, instead of type of condition (CL ± P and IH), type of cleft (cleft lip or cleft lip and palate) was included.

To account for multiple testing, *P* values were corrected using Bonferroni correction. All *P* values were 2 tailed. Statistical analyses were performed using IBM SPSS Statistics for Windows, version 25.0 (IBM Corp, 2017).

Results

A total of 337 families with children with CL ± P and 111 families with children with IH were eligible for the study, amounting to a total of 448 families. After invitation, 4 parents of children with CL ± P and 9 parents of children with IH indicated they did not have sufficient knowledge of the Dutch language. These parents were excluded from participation.

Of the families with a child with CL ± P, 173 mothers and 136 fathers returned completed questionnaires (46.3%). In the IH group, the response rate was 41.6% (53 mothers, 38 fathers). One mother in the CL ± P group had completed the questionnaires twice, so one duplicate record was removed prior to data analysis. A total of 400 parents (227 mothers, 173 fathers) from 237 families completed the questionnaires.

Preliminary Analysis

A nonresponse analysis did not reveal significant differences in age of the children with CL ± P between the participants and the nonresponders. However, on average, the children of nonresponders in the IH group were older (M = 4.55, SE = 0.58) than the children in the included IH sample (M = 3.59, SE = 0.38), $t(142) = -1.47$, $P < .05$.

Table 1. Sociodemographic Characteristics of the Sample.

	CL ± P, N = 309	IH, N = 91	*P* value
Parent gender, N (%)			
Male	136 (44.00)	38 (41.76)	.703
Female	173 (56.00)	53 (58.24)	
Parent age (years), mean (SD)	40.30 (6.31)	36.40 (5.73)	<.001
Parent age (years), range	25.58-71.42	24.00-49.92	
Child gender, N (%)			
Male	118 (65.19)	13 (23.21)	<.001
Female	63 (34.81)	43 (76.79)	
Child age (years), mean (SD)	7.23 (2.71)	4.30 (3.64)	<.001
Child age (years), range	2.42-12.50	0.08-11.67	
Nationality, N (%)			
Dutch	301 (97.40)	86 (94.51)	.181
Other/unknown	8 (2.60)	5 (5.49)	
Education, N (%)			
Low	34 (11.07)	9 (9.89)	.945
Average	146 (47.56)	44 (48.35)	
High	122 (39.74)	37 (40.66)	
Other/unknown	5 (1.62)	1 (1.10)	

Abbreviations: CL ± P, cleft lip with or without a cleft palate; IH, infantile hemangioma.

Of the 181 children with CL ± P, 66% of children had a cleft lip and palate and 34% had an isolated cleft lip. Of the 55 children with an IH, 90.1% of the children had an IH in the head and neck region. The other 8.9% had an IH on another location of the body. Prior to the main analysis, group differences in sociodemographic characteristics were tested. On average, parents of children with IH were younger than those of children with CL ± P, $t(160.46) = 5.57$, $P < .001$, and children with IH were younger than children with CL ± P, $t(121.24) = 7.93$, $P < .001$. Furthermore, there were more girls in the group of children with IH than in the CL ± P group, $\chi^2(1) = 41.45$, $P < .001$. Table 1 presents the families' sociodemographic characteristics.

Parents rated the child's condition as more visible for children with IH than children with CL ± P, $t(112.68) = -7.40$, $P < .001$. Parents of children with CL ± P rated their children as having more additional difficulties, $t(269.73) = 8.90$, $P < .001$, and as having more behavioral problems, $t(81.77) = 3.17$, $P = .002$, than parents of children with IH.

Distress and Parenting Stress Compared to Normative Data

As presented in Table 2, both mothers and fathers in the CL ± P and IH group had significantly less parenting stress than the normative population, with large effect sizes ($d = -0.79$ - $d = -1.53$). Furthermore, all parent groups scored significantly below the normative mean range, indicating low levels of parenting stress in parents of children with CL ± P or IH.

Table 2. One Sample *t*-test for Observed and Normative Mean Scores of Distress and Parenting Stress.

		Sample		Norm group			
Instrument	Condition	N	Mean (SD)	Mean (SD)	Mean range	*P* value	Cohen *d*
Fathers							
Parenting stress[b]	CL ± P	134	35.82 (15.41)	48.5 (16.4)	39-53	<.001[a]	−0.82
	IH	37	34.38 (17.97)	48.5 (16.4)	39-53	<.001[a]	−0.79
Anxiety[c]	CL ± P	134	11.39 (2.47)	12.23 (3.80)	12-14	<.001[a]	−0.34
	IH	38	12.16 (4.55)	12.23 (3.80)	12-14	.923	−0.02
Depression[d]	CL ± P	134	19.57 (6.30)	20.58 (6.76)	20-23	.067	−0.16
	IH	38	20.90 (9.37)	20.58 (6.76)	20-23	.834	0.03
Hostility[e]	CL ± P	134	7.19 (2.11)	7.22 (2.11)	7-8	.855	−0.02
	IH	37	7.58 (2.97)	7.22 (2.11)	7-8	.462	0.12
Mothers							
Parenting stress[b]	CL ± P	171	37.33 (17.33)	54.4 (19.3)	43-61	<.001[a]	−0.99
	IH	53	33.76 (13.50)	54.4 (19.3)	43-61	<.001[a]	−1.53
Anxiety[c]	CL ± P	170	12.04 (3.63)	13.43 (4.91)	12-14	<.001[a]	−0.38
	IH	53	11.91 (3.33)	13.43 (4.91)	12-14	.002[a]	−0.46
Depression[d]	CL ± P	169	21.33 (7.11)	22.89 (8.24)	20-23	.005	−0.22
	IH	53	21.09 (5.78)	22.89 (8.24)	20-23	.028	−0.31
Hostility[e]	CL ± P	169	7.19 (1.91)	7.33 (2.11)	7-8	.334	−0.07
	IH	53	7.11 (1.87)	7.33 (2.11)	7-8	.402	−0.12

Abbreviations: CL ± P, cleft lip with or without a cleft palate; IH, infantile hemangioma; Symptom Checklist – 90.
[a]Significant at a Bonferroni-corrected level of $\alpha = .003$.
[b]As measured by the NOSI-K, using norms of children aged 2 to 13 years.
[c]As measured by the SCL-90 Anxiety subscale.
[d]As measured by the SCL-90 Depression subscale.
[e]As measured by the SCL-90 Hostility subscale.

Both mothers and fathers of children with CL ± P reported less anxiety than the normative population, with small to medium effect sizes. For IH, only mothers reported significantly less anxiety (M = 11.91, SD = 3.33, $P = .002$, $d = -0.46$). Fathers of children with IH scored within the normal range, with a negligible effect size. On measures of depression and hostility, none of the parent groups differed significantly from the normative population.

Overall, when compared to the norm group, 3.49% of fathers and 3.08% of mothers were classified as scoring extremely high on parenting stress. Also, 4.05% of fathers and 2.20% of mothers scores were classified as high on parenting stress. For anxiety, 2.25% of parents scored extremely high and 9.25% parents scored above average/high. For depression, 4.25% of parents scored extremely high, while 11% scored high. For hostility, 5.25% of parents scored extremely high, while 8.75% parents scored high/above average. Detailed sample characteristics, split by parent gender and child condition, are summarized in Table 3.

Distress and Parenting Stress in Both Parent Groups

Linear mixed models were run to test our research question 2. Results are given in Table 4. All models had the lowest −2 log likelihood ratio, and thus the best fit, when including the covariates parent age, child age, child gender, child behavioral problems, visibility of the condition, and other problems related to the condition.

Regarding parenting stress, parents of children with CL ± P reported higher levels of parenting stress than parents in the IH group, $F(233.22) = 5.20$, $P = .023$. As noted earlier, these stress levels were lower than the normative group. Child behavioral problems, $F(286.50) = 191.54$, $P < .001$; child diagnosis, $F(233.22) = 5.20$, $P = .023$; and other condition-related problems, $F(295.32) = 14.60$, $P < .001$, were positively related to parenting stress. Parents of children with more behavior problems and other condition related had higher levels of parenting stress. Parents of children with CL ± P experienced more parenting stress than parents of children with an IH. Visibility of the condition was not related to parenting stress nor was parent age, parent gender, child age, or child gender.

Concerning feelings of anxiety, none of the independent variables reached significance. Feelings of anxiety were not related to the condition of the child (CL ± P or IH) nor to any of the proposed covariates.

The model for parental depression showed no significant effect of condition or children's age. Child behavioral problems, $F(1,182.36) = 9.61$, $P = .002$, and additional problems related to the medical condition, $F(328) = 9.85$, $P = .020$, were significantly related to parenting symptoms of depression, with parents of children with more behavioral and additional problems reporting more depressive symptoms.

Concerning hostility, condition (CL ± P or IH) did not significantly influence the model, but additional condition-related difficulties did, $F(254.82) = 8.73$, $P = .003$.

Table 3. Clinical Interpretation of Scores of Distress and Parenting Stress, N(%).

	Condition	N	Below average	Average	Above average[a]	High[a]	Extremely high
Fathers							
Parenting stress[b]	CL±P	133	98 (73.7)	22 (16.5)	3 (2.3)	6 (4.5)	4 (3.0)
	IH	37	29 (78.4)	3 (8.1)	3 (8.1)	2 (5.4)	3 (8.1)
Anxiety[c]	CL±P	134	95 (70.9)	30 (22.4)	7 (5.2)		2 (1.5)
	IH	38	26 (68.4)	7 (18.4)	4 (10.5)		1 (2.6)
Depression[d]	CL±P	134	96 (71.6)	21 (15.7)	4 (3.0)	9 (6.7)	4 (3.0)
	IH	37	23 (62.2)	8 (21.6)	3 (8.1)	1 (2.7)	2 (5.4)
Hostility[e]	CL±P	134	61 (45.5)	55 (41.0)	12 (9.0)		6 (4.5)
	IH	38	19 (50.0)	11 (28.9)	3 (7.9)		5 (13.2)
Mothers							
Parenting stress[b]	CL±P	169	130 (76.9)	25 (14.8)	3 (1.8)	5 (3.0)	6 (3.6)
	IH	53	42 (79.2)	8 (15.1)	2 (3.8)	0 (0)	1 (1.9)
Anxiety[c]	CL±P	171	110 (64.3)	37 (21.6)	19 (11.1)		5 (2.9)
	IH	53	38 (71.7)	7 (13.2)	6 (11.3)		2 (3.8)
Depression[d]	CL±P	171	92 (53.8)	36 (21.1)	5 (2.9)	29 (17.0)	9 (5.3)
	IH	53	28 (52.8)	15 (28.3)	2 (3.8)	5 (9.4)	3 (5.7)
Hostility[e]	CL±P	171	79 (46.2)	67 (39.2)	18 (10.5)		7 (4.1)
	IH	53	28 (52.8)	17 (32.1)	5 (9.4)		3 (5.7)

Abbreviations: CL±P, cleft lip with or without a cleft palate; IH, infantile hemangioma; SCL-90, Symptom Checklist – 90.
[a]For the anxiety and hostility scales, norm scores are reported in a combined category of above average and high scores in the manual.
[b]As measured by the NOSI-K, using norms of children aged 2 to 13 years.
[c]As measured by the SCL-90 Anxiety subscale.
[d]As measured by the SCL-90 Depression subscale.
[e]As measured by the SCL-90 Hostility subscale.

Table 4. Mixed Linear Models for Distress and Parenting Stress for Total Sample.

		Visibility	Condition[h]	Behavioral problems[i]	Condition-related problems	Parent age	Parent gender	Child age	Child gender
Parenting stress[d]									
B		.61	−4.84[a]	.52[c]	1.36[c]	.18	−1.69	.20	−.47
SE		0.42	2.08	0.04	0.37	0.12	1.10	0.29	1.50
95% CI	Lower	−0.20	−8.93	0.45	0.64	−0.07	−3.87	−0.38	−3.42
	Upper	1.43	−0.75	0.59	2.08	0.42	0.49	0.78	2.48
Anxiety[e]									
B		.10	−.43	.01	.19	<−.01	.36	.15	−.45
SE		0.11	0.53	0.01	0.10	0.03	0.34	0.08	0.38
95% CI	Lower	−0.12	−1.48	<−0.01	<−0.01	−0.06	−0.32	<−0.01	−1.20
	Upper	0.32	0.62	0.03	0.38	0.07	1.05	0.30	0.31
Depression[f]									
B		.30	−1.27	.06[b]	.49[a]	<−.01	.83	.29	−1.03
SE		0.23	1.09	0.02	0.20	0.07	0.74	0.16	0.78
95% CI	Lower	−0.15	−3.42	0.02	0.09	−0.14	−0.62	<−0.01	−2.56
	Upper	0.75	0.88	0.10	0.88	0.13	2.28	0.60	0.50
Hostility[g]									
B		.05	−.10	<.01	.19[b]	−.02	−.15	<−.01	−.07
SE		0.07	0.36	0.01	0.06	0.02	0.20	0.05	0.26
95% CI	Lower	−0.10	−0.81	−0.01	0.06	−0.06	−0.55	−0.10	−0.58
	Upper	0.19	0.61	0.02	0.31	0.02	0.25	0.10	0.44

Abbreviations: CL±P, cleft lip with or without a cleft palate; IH, infantile hemangioma; SCL-90, Symptom Checklist – 90.
[a]$P < .05$.
[b]$P < .01$.
[c]$P < .001$.
[d]As measured by the NOSI-K, using norms of children aged 2 to 13 years.
[e]As measured by the SCL-90 Anxiety subscale.
[f]As measured by the SCL-90 Depression subscale.
[g]As measured by the SCL-90 Hostility subscale.
[h]CL±P or IH.
[i]As measured by the CBCL using measures developed for children aged 1½ to 5 and 6 to 18.

Table 5. Mixed Linear Models for Distress and Parenting Stress for Parents of a Child With CL ± P.

		Visibility	Cleft type[h]	Behavioral problems[i]	Condition-related problems	Parent age	Parent gender	Child age	Child gender
Parenting stress[d]									
B		.66	.65	.49[c]	1.30[b]	.15	−.92	.23	−.61
SE		0.51	1.93	0.04	0.40	0.14	1.19	0.34	1.71
95% CI	Lower	−0.34	−3.16	0.41	0.51	−0.11	−3.27	−0.44	−3.99
	Upper	1.66	4.45	0.57	2.09	0.42	1.43	0.89	2.78
Anxiety[e]									
B		.12	−.36	.01	.17	−.01	.57	.18[a]	−.57
SE		0.14	0.49	0.01	0.11	0.04	0.38	0.09	0.42
95% CI	Lower	−0.15	−1.34	−0.01	−0.04	−0.08	−0.19	0.01	−1.43
	Upper	0.39	0.62	0.04	0.38	0.06	1.33	0.35	0.29
Depression[f]									
B		.34	.59	.05[a]	.51[a]	−.04	1.32	.35	−.62
SE		0.28	1.01	0.02	0.22	0.07	0.74	0.18	0.88
95% CI	Lower	−0.20	−1.41	<0.01	0.09	−0.18	−0.14	<−0.01	−2.37
	Upper	0.89	2.60	0.09	0.94	0.11	2.79	0.70	1.14
Hostility[g]									
B		.06	−.31	<.01	.15[a]	−.03	−.12	.04	−.11
SE		0.09	0.33	0.01	0.07	0.02	0.22	0.06	0.29
95% CI	Lower	−0.11	−0.96	−0.01	0.02	−0.08	−0.56	−0.08	−0.69
	Upper	0.24	0.34	0.02	0.29	0.01	0.31	0.15	0.47

Abbreviation: SCL-90, Symptom Checklist – 90.
[a]$P < .05$.
[b]$P < .01$.
[c]$P < .001$.
[d]As measured by the NOSI-K, using norms of children aged 2 to 13 years.
[e]As measured by the SCL-90 Anxiety subscale.
[f]As measured by the SCL-90 Depression subscale.
[g]As measured by the SCL-90 Hostility subscale.
[h]Cleft lip or cleft lip + palate.
[i]As measured by the CBCL using measures developed for children aged 1½ to 5 and 6 to 18.

Overall, none of the covariates (ie, parent age, parent gender, child age, child gender, or visibility of the condition) were significantly related to measures of either distress or parenting stress.

Secondary Analyses for Cleft Type

To assess whether cleft type influenced the amount of distress and parenting stress, mixed linear models were run with cleft type as a covariate. Results are provided in Table 5. Cleft type did not significantly influence the amount of parenting stress, anxiety, depression, or hostility in parents. Including cleft type in the models did not change the significance of predictors for parenting stress, depression, and hostility in comparison to the main analysis. For anxiety, child age was significantly related to parental stress, $F(50.83) = 4.64$, $P = .036$. Parents of older children experienced more anxiety than parents of younger children. This effect was not found in the main analyses.

Discussion

The present study aimed to determine whether parents of children with CL ± P or IH experience more distress and parenting stress than parents from the general population. Furthermore, we sought to understand how visibility and other parent-reported factors associated with the medical condition are related to distress and parenting stress. Our findings indicate that parents of children with a visible difference (CL ± P or IH) report lower levels of parenting stress and less symptoms of anxiety than parents from the general population. They did not differ significantly from the general population in feelings of depression and hostility.

Current results are not consistent with our expectations, as we expected parents of children with CL ± P and IH to report higher levels of distress and parenting stress. This expectation was primarily based on meta-analyses by Pinquart (2018a, 2018b, 2019) reporting elevated levels of distress and parenting stress in parents of children with a medical condition. An explanation for the current findings may be post-traumatic growth or a response shift.

Historically, the majority of research has focused on the negative impact on parents of having a child with a medical condition, resulting in poor adjustment, parenting stress, depression, or anxiety (Scorgie & Sobsey, 2000). However, there is a growing body of literature on stress-related growth and beneficial effects among parents of children with a medical condition (eg, Li et al., 2012). This is termed post-traumatic growth and is very common. About 58 to 83% of people who

have experienced a traumatic event report positive change in at least 1 life domain (Jayawickreme & Blackie, 2014). The present study shows that the parents of children with CL ± P or IH report less stress and equal levels of distress compared to adults in the general population. The current results can be seen in the light of post-traumatic growth, but more research is needed to study this phenomenon.

Another possibility is that a response shift occurs in parents of children with a medical condition. Response shift refers to changing internal standards, values, and the conceptualization of the target construct, such as quality of life (Sprangers & Schwartz, 1999; Schwartz et al., 2006). For example, research has shown that caregivers of patients with head and neck cancer experience fewer symptoms of depression and anxiety 6 months after diagnosis, compared to 0 or 3 months after diagnosis (Lee et al., 2017). Although longitudinal data are needed to detect a response shift, parents of children with CL ± P or IH may undergo a process of adaptation to the demands of having a child with a visible difference. Similarly, as included in the model by Wallander et al. (1989), it may be that parents in this sample have developed strong coping skills that helped mitigate possible risk factors.

Research question 2 focused on factors related to the levels of distress and parenting stress. We found that subjective visibility was not related to distress or parenting stress. On the other hand, child behavioral problems were significantly and positively related to parenting stress and parental depression. Furthermore, additional problems related to the medical condition were positively related to parenting stress, depression, and hostility. Correspondingly, parents of children with CL ± P reported more parenting stress than parents of children with an IH, possibly due to more additional condition-related problems. However, the type of cleft (cleft lip or cleft lip and palate) did not influence the amount of distress and parenting stress. Parent age, child age, and child gender were not significantly related to distress and parenting stress. When comparing the results to the model by Wallander et al. (1989), child behavioral problems and additional condition-related difficulties were risk factors for lower parental adaption, while the parent-reported degree of visibility of the condition was not related to parental adaptation.

Although the present study does support previous research showing that learning and feeding difficulties (Lockhart, 2003), behavioral problems and disease complexity (Pinquart, 2018b) are related to parental burden; however, several of our findings are inconsistent with those from previous studies. First, although the subjective visibility is generally seen as a consistent predictor of maladjustment for people with a visible difference (Moss, 2005), parental subjective visibility of the condition in their children was not related to any of the outcome variables in the present study. As this study concerns parents, who are not the direct focus of the onlookers' reactions, different effects might come into play. It may be that the subjective visibility of the condition is primarily limited to maladjustment in the person with the visible difference and not to maladjustment in family members.

Second, neither the children's age nor their parents' age was significantly related to distress or parenting stress. This is in contrast with Pinquart (2018b, 2019) who reported that parents of older children experience less parenting stress and anxiety than parents of younger children. However, this may be explained by the fact that Pinquart (2018b) included all medical conditions for chronically ill children, while the current study only looked at CL ± P and IH. In addition, the age of children in the sample skewed younger with a large group of infants with IH.

The result that child behavioral problems were associated with parenting stress and depressive symptoms is significant, as our sample primarily included children with few behavioral problems. The scores on the CBCL ranged from 0 to 111, with 75% scoring 30 or lower (van Dalen et al, manuscript submitted for publication). This might imply that even with relatively few behavioral problems, parents may experience more stress and depressive symptoms. This finding is consistent with earlier literature, indicating that child behavioral problems are related to parenting stress (Neece et al., 2012; Buodo et al., 2013). However, as previous literature has indicated a bidirectional relationship between child behavioral problems and parenting stress (Neece et al., 2012), the interplay between the 2 remains subject to debate.

This study also has some strengths and limitations. A particular strength of this study is the substantial number of parents (N = 400) participating in this study. Besides including 227 mothers, we were also able to include 173 fathers. As most studies often focus on mothers, we believe that this study makes an important contribution to current knowledge about fathers of children with CL ± P and IH, which was similar to the results found for mothers. We also contributed to the scarce IH literature by reporting on distress and parenting stress in parents of children with IH. Despite these strengths, the current study also has some limitations.

First, we used normative data with a large norm group representative of the population, which reduces bias. However, for the SCL-90, it is not clear how many people in this group are parents. This might hamper the comparison of our group of parents with the normative data. Second, the CBCL 1½-5 (Achenbach & Rescorla, 2000) and NOSI-K (de Brock et al., 1992) are appropriate for children aged 1.5 years and older and 2 years and older, respectively. By including children younger than these ages in the IH group reflecting the key phase of their medical monitoring and treatment, our results should be interpreted with caution. Third, the cross-sectional design of the current study limits the strengths of our conclusions. A longitudinal design could be more informative so that it is also possible to assess the development of distress and parenting stress over time. Fourth, this study only uses self-report measures. Assessing mental health in structured clinical interviews could contribute to assessing distress and parenting stress in a more detailed way.

We recommend that future research on distress and parenting stress assesses the influence of personal growth or positive adjustment on distress and parenting stress. More insight into

these factors may offer a more practical approach for clinicians in recognizing parents experiencing a burden when caring for their child with a visible difference.

In conclusion, parents of children with CL ± P or IH in this sample seem well adjusted to having a child with a visible difference. As some parents were experiencing significant difficulties, careful screening is needed to identify parents in need of support. Further, as child behavioral problems are related to both parenting stress and anxiety, we recommend monitoring parents of children with behavioral problems and, if indicated, providing appropriate psychological support.

Authors' Note

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