Stigmatization Predicts Psychological Adjustment and Quality of Life in Children and Adolescents With a Facial Difference

Ornella Masnari,1 MSc, Clemens Schiestl,2 MD, Jochen Rössler,3 MD, Stefanie K. Gütlein,3 MSc, Kathrin Neuhaus,2 MD, Lisa Weibel,4 MD, Martin Meuli,2 MD, and Markus A. Landolt,1 PhD

1 Department of Psychosomatics and Psychiatry, University Children’s Hospital Zurich, 2 Pediatric Burn Center, Plastic and Reconstructive Surgery, University Children’s Hospital Zurich, 3 Center for Pediatrics and Adolescent Medicine, University Medical Hospital Freiburg, and 4 Department of Pediatric Dermatology, University Children’s Hospital Zurich

All correspondence concerning this article should be addressed to Markus Landolt, Department of Psychosomatics and Psychiatry, University Children’s Hospital Zurich, Steinwiesstrasse 75, 8032 Zurich, Switzerland. E-mail: markus.landolt@kispi.uzh.ch

Received April 10, 2012; revisions received September 3, 2012; accepted September 5, 2012

Objectives This cross-sectional study assessed psychological adjustment and health-related quality of life (HRQOL) in children and adolescents with congenital or acquired facial differences and identified potential predictors of adjustment. Methods Data were obtained from 88 children, ages 9 months to 16 years, by means of parent questionnaires (n = 86) and standardized interviews with children ≥7 years old (n = 31). Evaluation measures included the Child Behavior Checklist (CBCL), KIDSCREEN-27, TNO-AZL Preschool Quality of Life Questionnaire (TAPQOL), and Perceived Stigmatization Questionnaire. Results Psychological adjustment, as measured by the CBCL, was within norms. Parent-reported HRQOL was good in preschool children. Parent- and self-reported HRQOL of participants 7–16 years old was impaired in several dimensions, including psychological well-being. Psychological adjustment (especially internalizing behavior problems) and HRQOL were predicted primarily by perceived stigmatization. Conclusions Identification of stigma experiences and appropriate support may be crucial to enhancing psychological adjustment and quality of life in children with facial disfigurement.

Facial differences can result from a wide range of conditions, including congenital malformations (e.g., cleft lip, port wine stains, nevi), injuries (e.g., burns), and dermatological diseases (e.g., psoriasis). Despite important advances in medical and surgical interventions, complete resolution of such conditions is rarely obtainable. Therefore, it is important to examine the consequences of a facial difference on the psychological adjustment and health-related quality of life (HRQOL) of affected individuals.

Facial appearance exerts a strong impact on social interaction and personal development (Langlois et al., 2000). Consequently, facial differences are presumed to negatively affect social encounters and to put individuals at risk for psychosocial difficulties and impaired HRQOL (Topolski, Edwards, & Patrick, 2005). Research findings confirm that individuals with visible differences are likely to experience stigmatizing behaviors, such as staring, avoiding, teasing, and manifestations of pity (Lawrence, Rosenberg, Mason, & Fauerbach, 2011; Masnari et al., in press; Strauss et al., 2007). Yet, data on the psychological adjustment and HRQOL of children and adolescents with visible differences are controversial. Several studies among individuals with various facial conditions have reported no major psychological maladjustment (Dieterich-Miller, 1992; Landolt, Grubenmann, & Meuli, 2000; Sheerin, MacLeod, & Kusumakar, 1995). However, there is some evidence of impaired HRQOL and difficulties in particular
areas of functioning, the most frequent of which relate to negative self-perceptions, emotional problems, and social functioning (Hunt, Burden, Hepper, & Johnston, 2005; Stubbs et al., 2011; Topolski et al., 2005). Previously suggested determinants of adjustment to visible differences in children include medical variables, characteristics of the child, and family/social variables. There is some evidence that acquired conditions are associated with more difficulties than congenital conditions (Patrick et al., 2007). It has been suggested that condition-specific effects are less important predictors of individual adjustment than one might expect (Rumsey and Harcourt 2007). Notably, numerous studies have shown that the severity of a condition is not a reliable predictor of psychological distress (Thompson & Kent, 2001). Findings about the impact of socioeconomic status (SES), age, and gender are inconsistent (Hunt et al., 2005). The quality of family relationships, parental adjustment, and social support have been found to be of great importance (Noronha & Faust, 2007). Teasing and bullying experiences have been shown to be negatively associated with mental health (Hunt, Burden, Hepper, Stevenson, & Johnston, 2007; Rimmer et al., 2007). Yet, to our knowledge, there is no quantitative evidence on the association between child adjustment and perceived stigmatization, which includes not only experiences of teasing, but also exposure to staring and other disrespectful behaviors.

Current data on psychological adjustment and HRQOL in children and adolescents with facial differences are limited in several ways. First, studies on appearance-altering conditions (e.g., burn scars, infantile hemangioma, and congenital melanocytic nevi) seldom differentiate between subjects with facial and nonfacial differences, thereby making specific conclusions difficult. Second, most of the existing research on facial disfigurement has focused on subjects with a cleft lip/palate or other severe craniofacial malformations. These conditions often involve functional impairment (e.g., speech or eating difficulties); thus, findings may not be generalized to conditions that cause only esthetic impairment (e.g., port wine stains). Finally, little data exist on predictors of adjustment specific to facial differences, and there is no quantitative evidence on the impact of perceived stigmatization.

The objectives of our study were twofold. First, we aimed to assess psychological adjustment and HRQOL in young people with facial differences exerting an esthetic but no functional impact. We expected to find good overall psychological adjustment and good overall HRQOL, albeit with some impairment in the social and emotional domains. Second, we aimed to examine the importance of medical, individual, and family-related predictors of adjustment to facial differences. Based on previous findings, we expected the size of the facial difference not to be predictive of adjustment. In contrast, we expected perceived stigmatization to be a significant predictor of child psychological adjustment and HRQOL.

Methods
Participants and Procedure
The data presented in this article are part of comprehensive data collection assessing the psychosocial impact of facial differences in children and adolescents. Data on the frequency of child- and parent-reported stigma experiences as well as predictors of perceived stigmatization are presented elsewhere (Masnari et al., 2012).

Participants were recruited among outpatients of University Children’s Hospital Zurich, Switzerland, and University Medical Hospital Freiburg, Germany. The study was approved by the local ethics committee. Families were eligible for the study if their child met the following criteria: (a) a visible facial difference (burn scar, infantile hemangioma, port wine stain, or congenital melanocytic nevus) with a current size ≥1 cm²; (b) age between 9 months and 16 years; (c) at least 6 months postaccident for burn patients; (d) no evidence of mental retardation; and (e) a good understanding of German. Based on the medical records of the two hospitals, 126 eligible families were identified and contacted by letter: nine could not be reached, 15 did not respond, 11 refused participation, and 3 were excluded from analyses owing to incomplete data. Thus, 88 families were included (response rate = 69.8%). Nonparticipants consisted mainly of children with infantile hemangiomas (n = 28) and were slightly younger than participants (Mage = 4.54 vs. 6.31 years; t = –2.37, p < .05).

Data were obtained by means of parent questionnaires and standardized interviews with children aged ≥7 years. Parent-provided proxy reports on their child’s psychological well-being and HRQOL as well as information on possible predictors, including sociodemographic and medical variables as well as self-reports on their own mental health. Children ≥7 years old provided self-reports of their HRQOL.

Parents provided written informed consent. Parents of children <7 years old received standardized questionnaires by mail. Parents of children ≥7 years old were asked to return an answer form indicating whether they agreed to participate in the study and whether their child was willing to take part in a face-to-face interview. These standardized interviews were conducted by the first author either at the child’s home or at the hospital. To ensure that children...
could express their own views openly, they were interviewed separately from their parents. Parent questionnaires were handed out after the child interview. These included three separate booklets: one asked parents to conjointly provide information on their child (proxy ratings); the two other booklets asked mothers and fathers to provide information separately on their own mental health. In two of the 34 cases involving a child ≥7 years old, only the child participated, whereas in three of these cases, only the parent responded. Consequently, 54 parent reports were obtained for preschool children, and 32 parent and 31 self-reports for school-age children. In 35 cases, proxy ratings were provided by the mother, in seven cases by the father, and in 37 cases by both parents conjointly. In seven cases, which parent filled out the proxy form was not indicated. If families did not respond to the initial study invitation or if questionnaires were not returned within 2 weeks, a reminder was sent by mail. After two subsequent weeks without notice, families were contacted by phone. There was no remuneration for participating in the study; but travel costs were reimbursed.

**Measures**

**Health-Related Quality of Life**

HRQOL for children ages 9 months to 6 years was assessed by parental report, using an authorized German version of the TNO-AZL Preschool Quality of Life Questionnaire (TAPQOL) (Fekkes, Bruil, & Vogels, 2004). HRQOL for participants ages 7–16 years was assessed using the German parent and child form of the KIDSCREEN-27 (Bisegger, Cloetta, & the European KIDSCREEN Group, 2005).

The TAPQOL is a well-validated, standardized and reliable measure that assesses proxy reports of HRQOL in preschool children. It consists of 43 items, classified into 12 syndrome scales, assessing four global dimensions of HRQOL: physical, social, cognitive, and emotional functioning. Three syndrome scales (social functioning, motor functioning, and communication) are applicable only to children ≥1.5 years of age. Following the TAPQOL protocol, parents were asked to note problems in any of the mentioned domains (e.g., *Has your child had stomachache or abdominal pain?*) and to rate their child’s well-being related to the specific problem (*At that time, my child felt fine, not so good, quite bad, bad*). All items had a recall period of 1 week. A description of the items can be found in Fekkes et al. (2000). Syndrome scales were transformed into a 0–100 scale. Higher scores indicate better HRQOL. To obtain a measure of overall HRQOL, we computed a total score by averaging the scores of the four global dimensions, which previously were computed as the average of all underlying syndrome scales. Norms were retrieved from the scale manual and were based on data from 251 parents of healthy Dutch children between the ages of 10 and 60 months (Fekkes et al., 2004). Internal consistency in this study was acceptable to good for the total score and most syndrome scales, except for the scales measuring lung, stomach, social, and anxiety problems, which revealed poor internal consistencies (Table II).

The KIDscreen-27 is a standardized multidimensional generic instrument designed to assess self- and proxy-reported HRQOL in children and adolescents 8–18 years old. The parent form was administered as a questionnaire and the child form as an interview. Validity and reliability of this instrument have been confirmed (Ravens-Sieberer et al., 2007). The questionnaire contains 27 items assessing five dimensions: physical well-being, psychological well-being, parent relations and autonomy, social support and peers, and school environment. The items assess either the frequency or the intensity of a behavior or a feeling on a 5-point Likert scale, over a recall period of 1 week. Following the Swiss manual (Bisegger et al., 2005), scale scores were transformed into *T* values based on reference data from a community sample of >1,600 Swiss children and parents. In our study, children 7 years old (n = 3) were thereby compared with normative data of 8–11 year olds. A total score was computed by averaging the *T* scores over the five global scales. Internal consistency in this study was acceptable to good for the total score and most subscales for both self- and proxy reports, except for the subscale “school environment” (Table II).

**Psychological Adjustment**

The Child Behavior Checklist (CBCL) is a widely used, well-validated, standardized measure assessing parental reports of a child’s psychological adjustment (Achenbach, 1991; Achenbach & Rescorla, 2000). Two authorized German versions of the CBCL were used: the CBCL/1.5-5 (Arbeitsgruppe Deutsche Child Behavior Checklist, 2002) for children ages 18 months to 4 years and the CBCL/4-18 (Steinhausen, Winkler Metzke, & Kannenberg, 1996) for children ages 4–16 years. Both instruments yield scores for two broadband scales (internalizing and externalizing behavior problems), and an overall total behavioral problems score. Higher scores indicate greater psychological maladjustment. *T* scores were derived based on normative data. For the CBCL/4-18, reference values were drawn from 1964 healthy Swiss children (Steinhausen et al., 1996). For the CBCL/1.5-5, no Swiss/German norms are available. Therefore, *T* scores were calculated based on a community sample of 700 healthy U.S. children (Achenbach & Rescorla, 2000). In the current study,
internal consistency for the internalizing (α = .73/.87),
externalizing (α = .90/.93), and total behavioral problems
scales (α = .92/.95) was acceptable to excellent for both
the CBCL/1.5-5 and the CBCL/4-18, respectively.

Perceived Stigmatization
Child stigma experiences were assessed via a German trans-
lation of the parent form of the Perceived Stigmatization
Questionnaire (PSQ) (Lawrence, Rosenberg, Rimmer,
Thombs, & Fauerbach, 2010). The translation procedure
followed published guidelines (Brislin, Lonner, &
Thorndike, 1973), including the use of independent
back-translation. The parent form of the PSQ asks parents
to rate how often their child experienced a variety of
stigmatizing behaviors commonly reported by people
with appearance distinctions. It contains 21 items classi-
fied into three factors: absence of friendly behavior, staring/
confused behavior, and hostile behavior. Answer choices
are on a 5-point Likert scale, ranging from 1 (never) to
5 (always), with a recall period of 1 year. A PSQ total
score is obtained by averaging over all items. Higher
scores indicate higher perceived stigmatization. A recent
study confirmed the good psychometric properties of this
instrument (Lawrence et al., 2010). In the current study,
internal consistency for the PSQ total score was good for
both self- (α = .81) and proxy reports (α = .88).

Size of the Facial Difference
As many participants in our study were outpatients not
regularly seen by a physician, we had no up-to-date med-
ical records for all of them. Therefore, we assessed the size
of the facial difference by parent estimate. Parents were
asked to draw the extent of their child’s facial difference
on a face template. In the two cases in which only the child
participated, this information was assessed by the first
author at the interview with the child. The size of the
facial difference was categorized into four groups, accord-
ing to the extent of the face affected by the condition: ≤5,
>5–25, >25–50, or >50%. The initial categorization was
performed by the first author who carried out the inter-
views with the patients. The second author assessed this
classification for 20 randomly selected participants. With
agreement of 95%, inter-rater reliability was excellent.

Mental Health of Parents
Mental health of mothers and fathers was assessed inde-
dependently with the Symptom Checklist-27 (SCL-27), a
well-validated multidimensional measure (Hardt, Egle,
Kappis, Hessel, & Brahler, 2004). The Global Severity
Index (GSI) was used as an indicator of mental health.
Higher scores indicate poorer mental health. The SCL-27
was filled out by 85 mothers and 78 fathers; in 78 cases,
data were available from both parents. Internal consistency
of the GSI was α = .89 for mothers and α = .70 for fathers.

Socioeconomic Status
SES was calculated as a sum score (range: 2–12) based on
paternal occupation and maternal education. Specific
examples of occupational and educational levels were
provided in a previous article (Largo, Molinari, Comenale,
Weber, & Duc, 1989). Occupational levels were assessed
on a 6-point scale ranging from 1 (occupations that do not
require any school qualifications or vocational training) to 6
(occupations that require a University degree). Accordingly,
education was categorized into six levels from 1 (did not
graduate from compulsory school) to 6 (University degree).
For mothers, level of education was used instead of occu-
pation because, in Switzerland, mothers of young children
often resign from their jobs after their child’s birth to stay
at home with their children. Three social classes were
defined as follows: scores 2–5 as lower SES, scores 6–9
as middle SES, and scores 10–12 as upper SES. For statisti-
cal analysis, we used the sum score. A similar measure
has been used before and has been shown to be a valid
measure of SES in the Swiss community (Landolt,
Buehlmann, Maag, & Schiestl, 2009).

Statistical Analyses
Data were analyzed using the statistical package PAWS for
Windows, release 18. All analyses were performed with
two-tailed tests and p < .05 considered significant. For cat-
egorical comparisons, we used χ²-tests. For comparisons of
continuous data, we used Student’s t-tests. The differences
between sample means and reference data were quantified
by calculating effect sizes (Cohen’s d; 0.2 small, 0.5
medium, 0.8 large effect size) (Cohen, 1988). To obtain
a comparable measure of parent-reported HRQOL for all
participants, the parent-reported TAPQOL and
KIDSCREEN-27 total scores were both transformed into
norm-based t scores and combined into a single variable:
the parent-reported HRQOL total score. Four linear regres-
sion models were generated using the parent-reported
HRQOL total score and the three CBCL scores (total, in-
ternal, and external behavioral problems score) as depend-
ent variables, all of which were normally distributed. The
predictors were entered hierarchically in blocks; within
blocks, variables were entered simultaneously. The four
variable blocks are as follows: (1) child age, child gender,
SES; (2) maternal and paternal GSI; (3) type (acquired vs.
congenital) and size of facial difference; and (4) perceived
stigmatization. We chose this method to ensure that the
effect of the medical variables and the perceived
stigmatization were controlled for the variance contributed by sociodemographic variables and parental mental health. Owing to the small sample size in self-reports (n = 31), we elected not to attempt regression analysis for self-reported HRQOL.

Results
Sample Characteristics
Sample characteristics are summarized in Table I for the overall sample, as well as for preschool and school-age children separately. The majority of the preschool children had a congenital condition, whereas almost 60% of the school-age children had a burn scar. Also, small facial differences, covering 5% or less of the face, were more frequent in the younger age group. Notably, in general, most of the patients included in this study had a facial difference affecting the skin only, without marked distortion of facial features and without functional impairment. Parent-perceived stigmatization of their child was significantly higher in the older age group. The mental health of parents and SES of families did not differ between the two age groups. Most families were from the middle or upper class.

Psychological Adjustment
Psychological adjustment, as measured by the CBCL, was well within norms. Parents in our sample (n = 72) reported their children to have no more internalizing (M = 49.32, SD = 10.45, p = .58, d = .07), externalizing (M = 49.82, SD = 10.92, p = .89, d = 0.02), or total behavior problems (M = 50.07, SD = 11.03, p = .96, d = 0.01) than a community sample.

Health-Related Quality of Life
Tables II and III show the mean scores of the HRQOL measures for our sample and the reference groups. Parents of children ages 9 months to 6 years did not report any impairment of their child’s HRQOL as measured by the TAPQOL. Indeed, they described their child as more active (lively/energetic) and having a better appetite than the reference group. Conversely, overall parent-reported HRQOL for patients 7–16 years old was impaired relative to community norms. Specifically, physical, psychological, and school functioning were significantly poorer, exhibiting small to moderate effect sizes. Parent relations and autonomy also were slightly impaired, albeit not significantly. Social support, however, was reported to be normal. Self-reports of HRQOL were within published norms, except for one dimension: children and adolescents with a facial difference reported poorer psychological well-being. Notably, social support was slightly better than in the community sample, although this effect did not reach the significance level.

Predictors of Psychological Adjustment and HRQOL
Table IV summarizes statistics for the four regression models predicting proxy-reported psychological adjustment and HRQOL. The selected predictors accounted for 24% of the variance in the CBCL total behavior problem score, 34% of the internalizing and 8% of the externalizing behavior problem score, and 32% of the HRQOL score. All models were statistically significant, except for the one predicting externalizing behavior problems (p = .14). Child age and gender did not significantly predict either outcome variable. SES status was a significant predictor of HRQOL. With the entry of the parental mental health indexes in Step 2, there was a significant increase of the amount of variance explained for all outcome variables. However, maternal and paternal mental health scores, separately, were not significant predictors of the dependent variables, except for the internalizing behavior problem score, which was significantly predicted by paternal mental health. Notably, medical variables (i.e., the type (acquired vs. congenital) and size of the facial difference), entered in Step 3, did not significantly predict any outcome variable. Conversely, perceived stigmatization, entered in Step 4, accounted for a significant portion of the variance in all outcome variables, except for the externalizing behavior problem score. Thus, children and adolescents experiencing high levels of stigmatization were at greatest risk of psychological maladjustment (especially internalizing behavior problems) and low HRQOL.

Discussion
The purposes of this study were to assess psychological adjustment and HRQOL in children and adolescents with various kinds of facial difference and to identify possible predictors of adjustment. In line with our first hypothesis, parents in our sample reported no significant psychological maladjustment of their child as measured with the CBCL. This result is consistent with previous findings in children with facial hemangiomas, burns, or port wine stains (Dieterich-Miller, 1992; Landolt et al., 2000; Sheerin et al., 1995). Although one could argue that generic measures are not able to capture the specific problems of individuals with facial differences, this result still suggests that the children in our sample might not suffer from any major psychological maladjustment.
With regard to HRQOL, our hypothesis that children with facial differences would fare worse than controls was supported for school-aged children, but not for preschool children. In preschool children, parent-reports of child HRQOL were well within norms. This result is in contrast with previous findings among children with facial burns (Stubbs et al., 2011) or infantile hemangiomas (Hoornweg, Grootenhuis, & van der Horst, 2009). Several factors may explain this discrepancy, including differences in the measures used or certain characteristics of the samples. Stubbs et al. (2011), for example, assessed HRQOL with a burn-specific questionnaire, which is possibly more sensitive to appearance-related difficulties than our questionnaire. Hoornweg et al. (2009) used the same measure as in our study, but 34% of the children in their sample had hemangioma-related complications (e.g., bleeding or impaired vision), which was not the case in our sample. This may explain the better HRQOL reports in our study. For patients ages 7–16 years, parents reported a significantly poorer overall HRQOL compared with reference data. Specifically, physical, psychological, and school functioning were impaired, with small to moderate effect sizes. With regard to self-reported HRQOL, children and adolescents in our sample exhibited good overall HRQOL, but diminished psychological well-being. Findings of impaired HRQOL in this age group are in line with previous research among individuals with different facial conditions (Stubbs et al., 2011; Topolski et al., 2005). Interestingly, in our sample, social support was normal in both self- and parent reports. This supports the observation that, although a facial difference may cause some difficulties in social encounters, it does not prevent children from having close friendships (Feragen, Kvalem, Rumsey, & Borge, 2010).

The different outcomes in the two age groups may be related to a number of factors. First, the two HRQOL measures used in this study could have assessed different aspects of quality of life. Second, different reference groups were used for the two age groups; this may have influenced the evaluation of outcomes. Third, the two subsamples differed regarding the type and size of the participants’ facial differences. However, as multivariate analysis
demonstrated that these variables were not predictive of HRQOL, this seems not to be a decisive factor. Finally, it could be speculated that the psychosocial impact of a facial difference increases with age. Our data suggest that school-age children are more at risk for experiencing social stigmatization than younger children. As several parents in our sample have commented, toddlers may be too young to be aware of their condition. Yet, problems may evolve in early school years when children increasingly engage in social comparisons with peers (Rumsey &

Table II. Sample Means and Reference Data for Health-Related Quality of Life in Preschool Children

<table>
<thead>
<tr>
<th>Measure</th>
<th>Sample</th>
<th>Reference group</th>
<th>Effect size</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>n</td>
<td>M</td>
<td>SD</td>
</tr>
<tr>
<td>TAPQOL parent form</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Physical functioning</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sleeping (α = .86)</td>
<td>54</td>
<td>82.21</td>
<td>18.51</td>
</tr>
<tr>
<td>Appetite (α = .74)</td>
<td>54</td>
<td>94.29</td>
<td>8.99</td>
</tr>
<tr>
<td>Lung problems (α = .51)</td>
<td>54</td>
<td>94.75</td>
<td>11.13</td>
</tr>
<tr>
<td>Stomach problems (α = .48)</td>
<td>54</td>
<td>91.82</td>
<td>13.10</td>
</tr>
<tr>
<td>Skin problems (α = .81)</td>
<td>54</td>
<td>91.20</td>
<td>13.00</td>
</tr>
<tr>
<td>Motor functioning (α = .67)</td>
<td>40</td>
<td>98.28</td>
<td>5.29</td>
</tr>
<tr>
<td>Social functioning</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Social functioning (α = .58)</td>
<td>40</td>
<td>94.58</td>
<td>11.56</td>
</tr>
<tr>
<td>Problem behavior (α = .90)</td>
<td>53</td>
<td>73.32</td>
<td>23.90</td>
</tr>
<tr>
<td>Cognitive functioning</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Communication (α = .82)</td>
<td>39</td>
<td>91.83</td>
<td>12.09</td>
</tr>
<tr>
<td>Emotional functioning</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Anxiety (α = .51)</td>
<td>52</td>
<td>83.65</td>
<td>17.30</td>
</tr>
<tr>
<td>Positive mood (α = .80)</td>
<td>53</td>
<td>97.80</td>
<td>8.67</td>
</tr>
<tr>
<td>Liveliness (α = 1.00)</td>
<td>53</td>
<td>100.00</td>
<td>0.00</td>
</tr>
<tr>
<td>Total score (α = .71)</td>
<td>53</td>
<td>89.28</td>
<td>7.95</td>
</tr>
</tbody>
</table>

Note. The scales "motor functioning," "social functioning," and "communication" are only relevant for children aged ≥18 months. 
α = Cronbach alpha.

Table III. Sample Means and Reference Data for Health-Related Quality of Life in School-Age Children

<table>
<thead>
<tr>
<th>Measure</th>
<th>Sample</th>
<th>Reference group</th>
<th>Effect size</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>n</td>
<td>M</td>
<td>SD</td>
</tr>
<tr>
<td>KIDSCREEN-27 parent form</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Physical well-being (α = .92)</td>
<td>32</td>
<td>46.56</td>
<td>15.72</td>
</tr>
<tr>
<td>Psychological well-being (α = .88)</td>
<td>32</td>
<td>45.83</td>
<td>12.21</td>
</tr>
<tr>
<td>Autonomy &amp; parents (α = .69)</td>
<td>32</td>
<td>49.41</td>
<td>11.46</td>
</tr>
<tr>
<td>Social support (α = .88)</td>
<td>32</td>
<td>50.25</td>
<td>12.66</td>
</tr>
<tr>
<td>School environment (α = .49)</td>
<td>31</td>
<td>49.01</td>
<td>6.60</td>
</tr>
<tr>
<td>Total score (α = .86)</td>
<td>31</td>
<td>48.75</td>
<td>9.21</td>
</tr>
<tr>
<td>KIDSCREEN-27 child form</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Physical well-being (α = .86)</td>
<td>31</td>
<td>51.16</td>
<td>12.45</td>
</tr>
<tr>
<td>Psychological well-being (α = .78)</td>
<td>31</td>
<td>49.07</td>
<td>9.93</td>
</tr>
<tr>
<td>Autonomy and parents (α = .66)</td>
<td>31</td>
<td>54.68</td>
<td>11.23</td>
</tr>
<tr>
<td>Social support (α = .69)</td>
<td>31</td>
<td>54.00</td>
<td>8.99</td>
</tr>
<tr>
<td>School environment (α = .44)</td>
<td>31</td>
<td>53.50</td>
<td>6.84</td>
</tr>
<tr>
<td>Total score (α = .65)</td>
<td>31</td>
<td>52.48</td>
<td>6.48</td>
</tr>
</tbody>
</table>

Note. α = Cronbach alpha.

The reference group consists of >1,600 Swiss children and parents (Bisegger et al., 2005).

One-sample t-test with normative data.
Problems also may arise in adolescence, when appearance, peer approval, and identity issues become paramount (Edwards et al., 2005). In addition, adolescents may increasingly have to deal with difficulties by themselves, whereas younger children may benefit from greater parental support. All this is likely to cause greater vulnerability in older children and adolescents than in preschool children. On the flip side, age also may confer adjustment benefits through the development of coping strategies over time (Thompson & Kent, 2001). Our data do not allow any conclusive explanation for the different outcomes in the two age groups; this issue requires further longitudinal research.

Multivariate analysis revealed that psychological adjustment and HRQOL were not predicted by child age and gender. But good HRQOL was associated with high SES, which is in line with previous findings (Bradley & Corwyn, 2002). The simultaneous entry of maternal and paternal mental health indexes into the regression model contributed significantly to the prediction of all outcome variables. However, maternal and paternal mental health as individual variables, were not significant predictors for the outcome variables, except that paternal mental health significantly predicted child internalizing behavior problems. In line with our hypothesis and previous findings (Thompson & Kent, 2001), the size and the type (congenital vs. acquired) of the facial difference did not significantly predict psychological adjustment or HRQOL. This supports the notion that all children who look different face similar problems, irrespective of their specific condition (Rumsey & Harcourt, 2007). Notably, global psychological adjustment and HRQOL were primarily predicted by perceived stigmatization. Particularly, perceived stigmatization predicted significantly internalizing, but not externalizing behavior problems. This fits well with the notion that children with visible differences are especially at risk for internalizing problems, like anxiety, depression, and social withdrawal. To date, literature concerning the association between stigmatization and adjustment to disfigurement has been predominantly of a theoretical nature and based on qualitative research. This is the first study to provide quantitative evidence on this association in young people with facial differences. Concordant findings have been documented in research among adults with psoriasis (Richards, Fortune, Griffiths, & Main, 2001). The strong association between stigmatization and adjustment may be explained by several mechanisms. First, through stigma experiences, individuals with a facial difference could conclude that they are deficient relative to their peers, which can lead to negative self-images and subsequent psychological difficulties (Hunt et al., 2007). Second, affected individuals could react with avoidance of potentially painful social encounters, which again, may constrain their psychosocial development (Kish & Lansdown, 2000). Third, teasing and poor peer acceptance may contribute to emotional problems, like an increased sense of loneliness and social isolation (Rumsey & Harcourt, 2007).

The strengths of the current study are its inclusion of children with a broad spectrum of ages and facial

### Table IV. Predictors of Parent-Reported Psychological Maladjustment and Health-Related Quality of Life

<table>
<thead>
<tr>
<th>Predictor</th>
<th>Total behavior problems</th>
<th>Internalizing behavior problems</th>
<th>Externalizing behavior problems</th>
<th>Parent-reported health-related quality of life (n = 75)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>$\Delta R^2$</td>
<td>$\beta$</td>
<td>$\Delta R^2$</td>
<td>$\beta$</td>
</tr>
<tr>
<td>Step 1</td>
<td>.05</td>
<td>.13*</td>
<td>.03</td>
<td>.10*</td>
</tr>
<tr>
<td>Child age</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Gender of child</td>
<td>.09</td>
<td>.12</td>
<td></td>
<td>.10</td>
</tr>
<tr>
<td>Socioeconomic status</td>
<td>.03</td>
<td></td>
<td></td>
<td>.08</td>
</tr>
<tr>
<td>Step 2</td>
<td>.16**</td>
<td>.19**</td>
<td>.10*</td>
<td>.09**</td>
</tr>
<tr>
<td>Mental health (GSI) of mother</td>
<td>.16</td>
<td>.16</td>
<td>.09</td>
<td>.18</td>
</tr>
<tr>
<td>Mental health (GSI) of father</td>
<td>.16</td>
<td>.26*</td>
<td>.18</td>
<td>.07</td>
</tr>
<tr>
<td>Step 3</td>
<td>.04</td>
<td>.01</td>
<td>.03</td>
<td>.01</td>
</tr>
<tr>
<td>Acquired vs. congenital condition</td>
<td>.25</td>
<td>.18</td>
<td>.23</td>
<td>.17</td>
</tr>
<tr>
<td>Size of facial difference</td>
<td>.08</td>
<td></td>
<td>.06</td>
<td>.21</td>
</tr>
<tr>
<td>Step 4</td>
<td>.09**</td>
<td>.09**</td>
<td>.04</td>
<td>.19***</td>
</tr>
<tr>
<td>Parent-perceived stigmatization</td>
<td>.36**</td>
<td>.35*</td>
<td>.23</td>
<td>.53***</td>
</tr>
<tr>
<td>Total $R^2$</td>
<td>.34**</td>
<td>.42**</td>
<td>.19</td>
<td>.40***</td>
</tr>
<tr>
<td>Total $R^2$ adjusted</td>
<td>.24**</td>
<td>.34**</td>
<td>.08</td>
<td>.32***</td>
</tr>
</tbody>
</table>

Note. *p < .05, **p < .01, ***p < .001
differences, high participation rate, use of well-validated, multidimensional and standardized measures with reference data, assessment of self- and proxy reports, inclusion of fathers, and adoption of multivariate statistics. Post hoc power analysis \( (\alpha = .05, \text{two-tailed}) \) using the G*power software (Faul, Erdfelder, Lang, & Buchner, 2007) indicated that for each \( t \)-tests comparing sample means with normative data, the power to detect a large effect size \( (d = 0.8) \) exceeded .99. The power to detect a medium effect size \( (d = 0.5) \) was adequate for the CBCL (.99) and the TAPQOL (.83), but just below the recommended .80 level (Cohen, 1988) for the KIDSCREEN-27 (.77). The power to detect a small effect size \( (d = 0.2) \) was less than adequate for all outcome variables. For the regression analyses, the power to detect a large \( (f^2 = 0.35) \) or a medium \( (f^2 = 0.15) \) effect size was more than adequate (i.e., >.80) in each analysis, but the power to detect small-sized effects \( (f^2 = 0.02) \) was less than adequate. Thus, overall, our sample sizes provided adequate power at the medium to large effect size level, but not enough power at the small effect size level.

Further limitations to the present study exist. First, the cross-sectional design of this study prevents any conclusions about causal relations. Second, the exclusion of non-German-speaking families resulted in an underrepresentation of lower class families. Third, the appropriateness of using U.S. norms for the CBCL/1-5 and Dutch norms for the TAPQOL can be questioned. Slight differences in the age ranges of the normative samples and cultural differences may compromise comparability. This being said, cross-cultural bias seems unlikely, given previous findings supporting the use of U.S. norms for the CBCL in German samples (Elting, 2003), as well as similar HRQOL scores of chronically ill children within central European countries (Schmidt et al., 2006). Fourth, some subscales of the HRQOL measures exhibited low internal consistency. This raises doubts on the usefulness of these subscales in future research and calls for a reevaluation of their validity. However, the low internal consistency of some subscales did not compromise our multivariate analyses, for which we only used global HRQOL scores with good internal consistency. Fifth, the size of the facial difference was estimated based on parent perception, though categorized by raters; future research should try to implement a standardized measurement. Sixth, in multivariate analyses the use of both parent-reported predictors and outcome variables did not control for common source and method variance as factors that might contribute to significant findings. Finally, we elected not to attempt multivariate analysis with self-reported HRQOL, because of the small sample size for self-reports \( (n = 31) \); this issue warrants further investigation.

Future research activities should include longitudinal studies to disentangle the mechanisms behind the association between stigmatization and psychological maladjustment. Moreover, as the factors included in this study explained <30% of the variability in child adjustment, it will be important to examine other possible predictors, like the characteristics of family communication, child personality, or coping strategies. Notably, the question arises whether social support and good social skills might act as protective factors against the potential challenges of stigmatization. Finally, as generic measures may lack sensitivity for specific difficulties, condition-specific measures could be vital to studying the impact of facial disfigurement (Edwards et al., 2005; Patrick et al., 2007).

Regarding the clinical assistance of children with facial differences, corrective surgery may offer psychosocial benefits (Horlock, Voegelin, Bradbury, Grobbelaar, & Gault, 2005). But because a complete resolution of a facial difference is rarely obtainable, medical care should be accompanied by psychological assistance. Early identification of stigma experiences and appropriate support might be crucial to enhance psychosocial adjustment and quality of life among young people with facial differences. A combination of cognitive behavioral therapy and social skills training, with inclusion of the parents, (Kish & Lansdown, 2000) could be a particularly promising approach to assisting children and adolescents with visible facial differences.

**Acknowledgments**

The authors acknowledge the kind cooperation of all parents and children who participated in this study.

**Conflicts of interest:** None declared.

**References**


