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Coping and Family Functioning Predict Longitudinal Psychological Adaptation of Siblings of Childhood Cancer Patients

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Objective To assess associations of coping and family functioning with psychosocial adjustment in siblings of pediatric cancer patients at 1, 6, 12, and 24 months after diagnosis.

Methods Eighty-three siblings (ages 7–19 years) participated. Effects on anxiety, quality of life, behavioral-emotional problems, and emotional reactions to the illness were investigated. Data-analysis was performed with multilevel mixed modeling.

Results Psychosocial functioning was impaired at 1 month but ameliorated over time. Adjustment problems were associated with high family adaptation and cohesion, older age, and female gender. Lower anxiety, insecurity, loneliness, and illness involvement were related to siblings' ability to remain optimistic. Insecurity and illness involvement were positively related to reliance on the medical specialist and a tendency to seek information about the illness. **Conclusions** Siblings of pediatric cancer patients are most affected by the illness in the first months. Children at risk may be identified according to sibling age and gender and according to long-term family adaptation processes and sibling coping abilities.

Key words siblings; childhood cancer; adaptation; coping; family system.

As medical treatment of pediatric chronic or life-threatening diseases has improved and more and more children survive, the physical and psychosocial consequences of such treatment have become increasingly relevant in pediatric health care. Psychological consequences of chronic or life-threatening illness in childhood do not concern only the patient but also extend to family members (Kazak, 1989). Siblings of a child with cancer have been reported to suffer from the intensive treatment and the changes it brings about in their daily routines, emotional state, social life, and, of course, family relations. Internalizing (emotional) problems as well as externalizing (behavioral) problems have been reported in several studies (Barbarin et al., 1995; Bendor, 1990; Carpenter & Sahler, 1991; Cohen, Friedrich, Jaworski, Copeland, & Pendergrass, 1994; Fife, Norton, & Groom, 1987; Heffernan & Zanelli, 1997; Packman et al., 1997; Schuler et al., 1985; Spinetta, 1981; Walker, 1988). In a meta-

analysis of the literature on the psychological effects of a chronic or life-threatening illness on siblings, internalizing problems appeared to be the most prominent (Sharpe & Rossiter, 2002). Siblings of children with cancer experience feelings of isolation and anxiety (Cairns, Clark, Smith, & Lansky, 1979; Bendor, 1990), withdrawal (Carpenter & Sahler, 1991; Heffernan & Zanelli, 1997), jealousy, conflicting feelings such as guilt and anger, and loneliness (Chesler, Allswede, & Barbarin, 1992; Gogan & Slavin, 1981; Martinson, Gilliss, Colaizzo, Freeman, & Bossert, 1990). When more time elapses, mediating factors influencing the sibling's adjustment to the illness come into play, such as the course of the illness, coping behavior, family functioning and resources, and parent mental health. As many have commented before (Chesler et al., 1992; Madan-Swain, Sexson, Brown, & Ragab, 1993; Sloper & While, 1996; Sourkes, 1980; Walker, 1990), the focus of research has been on

siblings' adjustment outcomes and not so much on the processes that underlie a positive or negative outcome, such as coping and family functioning. Although many researchers have emphasized the necessity of longitudinal research on sibling adjustment (Carpenter & Sahler, 1991; Cohen et al., 1994; Kazak, 1989; Madan-Swain et al., 1993; Sloper & While, 1996), studies on sibling adjustment are still predominantly cross-sectional (Houtzager, Grootenhuis, & Last, 1999). However, the distinction between stressors on the one hand and distress responses on the other is hard to achieve when focusing on one cross-sectional time point (McCubbin et al., 1980). Cross-sectional studies do no justice to the dynamic nature of the illness and the adaptation process. They provide no information on which problems are acute and which problems fade, and do not allow us to control for factors that become apparent only as more time elapses. The importance of longitudinal research on sibling adjustment is demonstrated by Alderfer, Labay, and Kazak (2003), who observed posttraumatic stress reactions in almost 30% of 99 siblings of childhood cancer survivors. In the present study, therefore, the social-emotional adjustment of siblings to pediatric cancer in a brother or sister will be studied over time in association with several potential mediating factors: demographic characteristics of the sibling, illness characteristics, coping, and family functioning.

Although such information is needed in order to develop useful intervention programs, the effectiveness of coping strategies has rarely been investigated systematically in siblings of children with cancer using standardized instruments (Houtzager et al., 1999). Several researchers have commented that more sensitive measures of coping are needed in order to find meaningful results regarding effective coping (Houtzager et al., 1999; Sloper & While, 1996) and that a theoretical framework is lacking in many cases. The process model of Lazarus and Folkman (1984) has been the basis of most studies on coping and adjustment to illness. These authors define the coping process as comprising both cognitive and behavioral efforts to manage specific external and/or internal demands that are appraised as taxing or exceeding the individual's resources. An individual can undertake actions to solve the problem that causes distress (behavior focus) or try to change cognitions regarding the stressful situation in order to reduce negative emotions that result from it (emotion focus). The nature of the stressful situation can demand a certain way of coping (Kliewer & Sandler, 1992). When a family is confronted with the life-threatening illness of a child, there is little its members can do to change the situation or exert

direct control (behavioral or problem-focused coping). Siblings are particularly helpless because they are least of all directly or actively involved in the treatment process. Without control, they may have to rely primarily on cognitive or emotion-focused coping strategies. They can try to rely on the medical specialists' competence and keep faith in the treatment regimen, to remain optimistic and wish for better times, or to understand the situation in order to gain a sense of control. Grootenhuis and Last (2001) and Grootenhuis, Last, De Graaf-Nijkerk, and Wel (1996) have developed questionnaires to assess cognitive coping strategies in young cancer patients and their parents. The cognitive coping strategies investigated were based on the two-process model of perceived control by Rothbaum et al. (1982). Cognitive coping strategies revert to secondary control strategies or attempts to bring oneself in line with the situational demands. Grootenhuis and Last (2001) found a relationship between these coping strategies and the patients' emotional well-being. Houtzager, Grootenhuis, Hoekstra-Weebers, and Last (2003) found that these coping strategies were relevant for siblings of cancer patients as well and that cognitive coping strategies predicted the adjustment of siblings of pediatric cancer patients at 1 month after diagnosis. Still, it is important to know whether and which coping strategies are relevant throughout the treatment process.

Because childhood cancer can be extremely disruptive of family daily life and emotional well-being, it affects all family members. Therefore, a family-systems approach to the investigation of the impact of the disease on siblings is essential. Several researchers have reported increased closeness in families of a child with cancer (Chesler et al., 1992; Gogan & Slavin, 1981; Kramer, 1984; Sargent et al., 1995). Cohesiveness can reach levels that would normally be regarded as pathological, comparable to a so-called enmeshed family structure. This high interrelatedness may be regarded as a family coping strategy (Carpenter & Levant, 1994; Houtzager et al., 1999; Kazak, 1989). High family cohesion and adaptability have been found to relate to fewer adjustment problems in siblings (Cohen et al., 1994; Horwitz & Kazak, 1990). Inversely, problematic interpersonal or family relations appear to be related to more adjustment problems (Carpenter & Sahler, 1991; Fife et al., 1987; Sloper & While, 1996). The amount of distress experienced by parents may determine the amount of time and energy they have left for the sibling in the family and may therefore determine the sibling's well-being. Cohen et al. (1994) found a positive relationship between parental depressive symptoms and parent-reported adjustment

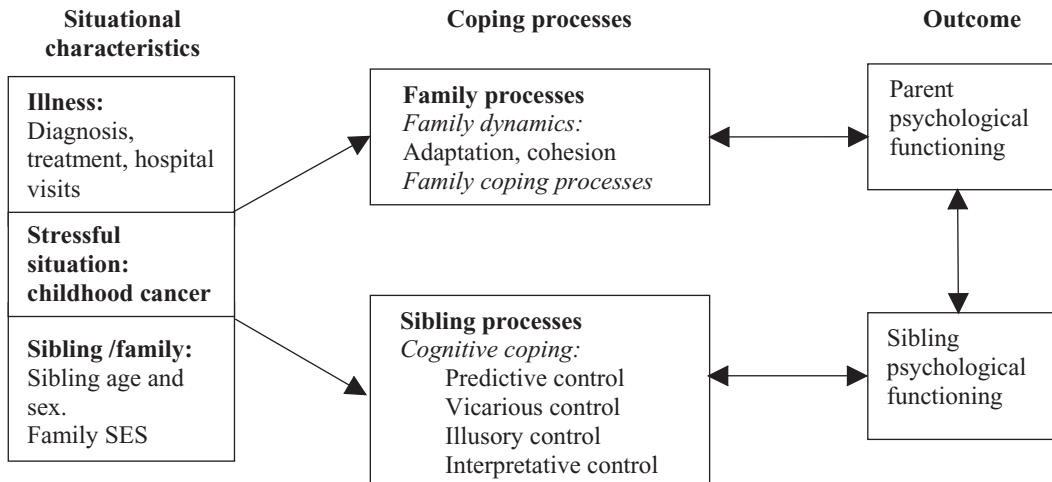


Figure 1. The adaptation processes: Situational characteristics, coping processes, and outcome.

problems in siblings. Sloper and While (1996), however, found no significant relationship between parental emotional distress and parent- and teacher-reported adjustment problems in 99 siblings 6 months after diagnosis. They argue, however, that parental distress may become relevant when it is of longer duration. Williams and colleagues (2002) demonstrated the complexity of the impact of parent mental health on sibling adjustment to chronic illness. Maternal mood had an indirect impact on sibling adjustment through its influence on family cohesion, sibling-experienced social support, and the amount of self-esteem a sibling had.

In the present study, a process- and family-oriented approach was applied to investigate the association of coping and family functioning with sibling psychological well-being over time, controlling for illness and sibling characteristics (Figure 1).

The main research question to be answered was, to what extent are cognitive coping strategies and family cohesion and adaptation related to the psychological well-being of siblings of pediatric cancer patients during the first 2 years following the diagnosis of cancer? Self-reported anxiety, emotional-behavioral problems, quality of life, and illness-specific emotional reactions were the domains of psychological well-being, which were assessed as dependent outcome variables. Besides, parent-proxy-reported psychological well-being was to be assessed as well. Sibling cognitive coping strategies, family adaptability and cohesion, and parent psychological distress were the independent mediating variables, which were assessed over time. Furthermore, associations of the independent variables with sibling psychological well-being were controlled for effects of gender, age, and illness characteristics.

Methods

The children and their parents participated in a longitudinal follow-up study of psychosocial adjustment in siblings of pediatric cancer patients. The study consisted of four assessments, at approximately 1 month (M1), 6 months (M2), 12 months (M3), and 24 months (M4) after the diagnosis of cancer in a brother or sister.

Participants

Seventy-one families were approached at 1 month after the diagnosis in the ill child. Informed consent was obtained from 56 families (78.9%) with 83 siblings (Table I). Families that refused participation did not differ significantly from the participants regarding sibling age ($t = 0.63, p = .53$) and sex ($\chi^2 = 0.38, p = .54$), nor did they differ in age ($t = -0.70, p = .49$) and sex ($\chi^2 = 0.09, p = .77$) of the ill child and type of diagnosis of the ill child ($\chi^2 = 1.70, p = .63$).

The study group consisted of 45 families (82%) and 66 siblings (80%) in the second measurement (M2), 40 families (71%) and 60 siblings (72%) in the third measurement (M3), and 38 families (68%) and 57 siblings (69%) in the last measurement (M4) (Table I). In the majority (61%) of the 18 families that dropped out of the study, the ill child had died. The other dropout families reported that they did not want to burden the family with participation or had other, unspecified reasons for discontinuation. The majority of the ill children in the dropout group had been diagnosed with a solid tumor or a brain tumor ($n = 14$) ($\chi^2 = 6.3, p = .01$), and brothers were relatively overrepresented compared with participants (65.4%) ($\chi^2 = 6.6, p = .01$). There was no difference in sibling age between groups. We compared

Table I. Characteristics (Sibling, Ill Child)

Measurement		M1	M2	M3	M4	
Time Since Diagnosis		1 Month	6 Months	12 Months	24 Months	
Siblings	N	83	66	60	57	
	Sex, boys, n (%)	37 (44.6)	26 (39.4)	25 (41.7)	20 (35.1)	
	Age, mean (SD)	(range: 7–19) 11.0 (2.8)	11.6 (2.8)	11.9 (2.8)	12.7 (2.8)	
	Age group, y, n (%)	7–11	48 (57.8)	38 (57.6)	32 (53.3)	23 (40.4)
		12–18	35 (42.2)	28 (42.4)	28 (45.2)	30 (52.6)
		>18				4 (7.0)
	Sibling older than ill child, n (%)	56 (67.5)	45 (68.2)	41 (68.3)	39 (68.4)	
Ill Child	N	56	45	40	38	
	Diagnosis, n (%)					
		Leukemia	12 (21.4)	11 (24.4)	11 (27.5)	12 (31.6)
		Lymphoma	13 (23.2)	11 (24.4)	11 (27.5)	10 (26.3)
		Solid tumor	26 (46.4)	21 (46.7)	17 (42.5)	15 (39.5)
		Brain tumor	5 (8.9)	2 (4.4)	1 (2.5)	1 (2.6)
	Days in hospital between assessments, mean (SD)	21.7 (10.0)	26.1 (20.6)	12.8 (23.3)	3.8 (6.6)	
	Sex, boys, n (%)	29 (51.8)	22 (48.9)	19 (46.3)	17 (44.7)	
	Both parents born in Netherlands, n (%)	44 (78.6)	38 (84.4)	34 (85.0)	33 (86.8)	

mean outcome scores at M1 of nondropouts and siblings in the 18 dropout families in order to account for selective dropout. No significant differences were found on any of the outcome measures that were investigated here.

Procedure

Siblings of children with cancer and their parents were recruited from the Emma Children's Hospital in the Academic Medical Center in Amsterdam and from the University Hospital in Groningen, the Netherlands. One or two siblings were included per family, in order to prevent overrepresentation of large families. Inclusion criteria were that siblings be 7–18 years at the time of the diagnosis, that the ill child have a first diagnosis of malignant pediatric cancer, and that siblings and parents be in sufficient command of the Dutch language.

Families were informed about the study by letter 3 to 4 weeks after the ill child was diagnosed with cancer. They were telephoned after 2 weeks or after informed consent had been returned by mail. An appointment was made for an assessment at the family's home if informed consent was obtained. Two psychologists conducted interviews with one or both parents and the sibling(s). The questionnaires were sent in advance, with explicit instructions to parent and child to complete them independently. Questionnaires were collected after the interviews. Approval of this study was obtained from the medical ethical committee of the Academic Medical Center in Amsterdam and Groningen.

Measures

Dependent Variables

Eight outcome variables were assessed: anxiety, quality of life, behavioral-emotional problems, and four illness-specific emotional reactions in siblings.

Anxiety. The State-Trait Anxiety Inventory for Children (STAI-C) aged 8–15 years (Spielberger, Edwards, Lushene, Montouri, & Platzek, 1973; Spielberger, Gorsuch, & Lushene, 1970) was used in the Dutch version (the Zelf-Beoordelings Vragenlijst voor Kinderen) (Bakker, Wieringen, Ploeg, & Spielberger, 1989). The trait version was used to assess the tendency to respond with anxiety in a threatening situation. In contrast to the state version of the STAI-C, which measures conditional anxiety at the very moment of assessment, the trait version is more appropriate for measuring the overall level of anxiety a child experiences. Scores on the STAI-C range from 20 to 60, with high scores representing high levels of anxiety. Reliability of the STAI-C in a Dutch population ranged from .81 to .88 (Bakker et al., 1989).

Quality of Life. The Dutch Children's AZL/TNO* quality of Life questionnaire (DuCATQoL) (Kolsteren, Koopman, Schalekamp, & Mearin, 2001) was used to measure the evaluation of the quality of daily functioning by children aged 7–15 years. The short form of the DuCATQoL consists of 25 items, scored on a 5-point scale. It can be used to assess the affective appraisal of

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daily functioning in children and consists of four domains: family functioning, bodily functioning, emotional functioning, and social functioning. The total quality of life (QoL) score that can be obtained from these scales will be reported here. High scores represent good QoL. Internal consistency (Cronbach's α) of the DuCATQoL was .85 in our study group at M1. Although no validation studies have been published yet, the DuCATQoL is reported to be internally consistent and reproducible (Kolsteren et al., 2001). Reference data for the DuCATQoL were available. These were obtained from a Dutch sample of 1,092 healthy children 8 to 12 years old, and 267 teenagers 12 to 15. These children were randomly selected by 12 municipal health services in the Netherlands, stratified by gender and age.

Behavioral-Emotional Problems. The Dutch Child Behavior Checklist (CBCL, ages 4–18 y) for parents and the Youth Self-Report (YSR, ages 11–18 y) (Achenbach & Edelbrock, 1983; Verhulst, Ende, & Koot, 1997) were used to assess the prevalence of behavioral and emotional problems as reported by parent and child, respectively. The CBCL was administered to parents of all siblings aged 7–18. The YSR was administered to siblings aged 11–18. Total behavior problem scores of both measures were used, comprising internalizing, externalizing, and other problems. Internal consistency (Cronbach's α) was high in the Dutch reference groups for the CBCL (.78–.92) and the YSR (> 0.85). Verhulst and colleagues (1997) assessed the 2-year stability of CBCL scores in a nonclinical norm population. Pearson's correlation coefficients were .65–.69 for the CBCL for parents and .59–.63 for the YSR.

Emotional Reactions. The Situation-Specific Emotional Reactions Questionnaire for siblings (SSERQ-s) was used to assess emotional reactions specific to siblings of a child with cancer that are not assessed with generic questionnaires. The SSERQ was originally developed to assess emotional reactions in parents of children with cancer (Grootenhuis & Last, 1997). A sibling version was based on the parent questionnaire and was adapted to the situation-specific emotions that siblings encounter, based on the literature. It was tested in a pilot study population of siblings and adapted according to reliability analyses and factor analyses. Factor analysis at M1 resulted in four different emotional domains: uncertainty, loneliness, emotional involvement, and positive emotions. Feelings of uncertainty consisted of eight items, for example *I am afraid that my brother or sister will not get well* and *I worry about the future*. The loneliness domain consisted of seven items, such as *I feel like I can talk to no one* and *I feel lonely*. The emotional-involvement domain

consisted of seven items, for example *I am sad because by brother/sister has to undergo painful procedures* and *I regret that my parents have to go through all this*. The domain for positive emotions consisted of three items, for example *I am proud that I can keep up with it*. Reliabilities at M1 were .84 for uncertainty, .79 for loneliness, .85 for emotional involvement, and .72 for positive emotions (Cronbach's α). Being an illness-specific questionnaire, no reference data are available.

Independent Variables

Coping, family functioning, and parent psychological distress were assessed as predictor variables for the siblings' adjustment, as were sibling and illness characteristics.

Coping. The degree to which siblings relied on cognitive coping strategies was measured with the Cognitive Coping Strategies Scale for siblings (CCSS-s). The CCSS-s is an illness-specific self-report questionnaire based on the original version developed for children with a chronic or life-threatening disease. The CCSS is used to assess to what extent children try to gain a sense of control over their illness by using cognitive coping strategies (Grootenhuis & Last, 2001). The CCSS-s consists of 20 statements with which children can indicate on a 4-point Likert scale to what extent they agree (1 = totally agree, 2 = agree, 3 = disagree, 4 = totally disagree). Four items were omitted after factor analysis. The questionnaire consists of scales for predictive control (optimism, efforts to maintain positive expectations regarding the illness; four items), vicarious control (putting trust in the treatment and medical staff or crediting them with positive characteristics; six items), interpretative control (efforts to gain understanding of the illness; four items), and illusory control (wishful thinking; two items). Internal consistency (Cronbach's α) was .81, .70, .71, and .30, respectively. Low internal consistency of the illusory-control scale indicates that this coping strategy did not sufficiently apply to siblings of children with cancer, and it was therefore omitted in the final analyses.

Family Functioning. The Dutch version of the Family Adaptability and Cohesion Evaluation Scales (FACES) (Buurmeijer & Hermans, 1988), originally developed by Olson and colleagues (Olson, Bell, & Portner, 1978; Olson, Portner, & Bell, 1982; Olson, Portner, & Lavee, 1985), was used to assess family functioning. Scores on family cohesion and adaptation can be obtained. The cohesion scale refers to mutual connectedness among family members and consists of 23 items. The adaptation domain refers to the level to which a family adapts its power structure, role definitions, and rules

according to internal and external demands, consisting of 13 items. Items are scored on a 4-point Likert scale (1 = never true, 2 = sometimes true, 3 = mostly true, 4 = always true). Cronbach's- α reliability of the Dutch FACES is good: .87 for cohesion and .81 for adaptation.

Parent Psychological Distress. The Dutch version of the 30-item General Health Questionnaire (GHQ-30) was administered to the mothers (Koeter & Ormel, 1991). In two cases, the fathers completed this questionnaire because the mothers were not available. The bimodal scoring recommended by Goldberg was applied to compute the total problem score, where 0 referred to symptoms present *less than usual* or *as usual*, and 1 referred to symptoms present *more than usual* or *much more than usual*. High scores represented high emotional distress. Reliability of the GHQ-30 is .93 (Cronbach's α) in the general Dutch population (Koeter & Ormel, 1991).

Sibling and Illness Characteristics. These included sibling age and gender, the ill child's diagnosis, and the number of hospital days between measurement occasions. Illness characteristics were obtained from the medical record of the ill child. The ill child's diagnosis was dichotomized into 1 = leukemia/lymphoma or 0 = a solid or brain tumor. Although siblings whose ill brother or sister died during the study did not continue participation, their psychosocial functioning assessments on previous measurement occasions were included in the analyses. To account for the larger likelihood of sibling distress during the more complex and serious disease process, eventual death of the ill child was included as another explanatory variable.

Statistical Analyses

Descriptive statistics were used to delineate the characteristics of the ill child and his or her siblings. Student *t* tests were used to compare siblings' mean scores on the standard questionnaires (STAI-C, DuCATQoL, CBCL, and YSR) with reference data.

The primary research questions were investigated through multilevel modeling. In this analysis, to facilitate interpretation of regression coefficients, all continuous scores on outcome and predictor variables were transformed into standard normal scores (with an overall mean of zero and an overall standard deviation of 1). Measurement occasions (level 1) were treated as nested within siblings (level 2), and siblings as nested within families (level 3). In this way dependencies between siblings that came from the same family were accounted for. The major advantage of multilevel analysis of longitudinal data is that all available data are incorporated

into the analysis, including data from families that missed one or more measurement occasions. Efficient estimates can be obtained through maximum likelihood estimation procedures if dropout is random. Analyses were carried out with Statistical Package for the Social Sciences version 11.5 (SPSS Inc., 2003) and MLwiN (Rasbash et al., 2000).

Three-level models were fitted for each of the eight outcome variables separately. Measurement occasions were treated as fixed (Snijders & Bosker, 1999). We also considered variable occasion models (various growth curve models), but these did not fit as well as the fixed occasion models according to chi-square tests of the improvement of fit over models with a random intercept only. Each model consisted of 15 regression coefficients, representing the deviations from the overall mean at M1, M2, M3, and M4; the effects of the diagnosis of the ill child; the number of hospital admission days; whether or not the ill child died during the study; the sibling's sex and age (at M1); the sibling's predictive, vicarious, and interpretative control; sibling-reported family cohesion and adaptation; and parent's psychological distress. The intercept was considered random, with its mean fixed at zero (i.e., constraining both fixed and random effects to zero, thus taking the overall mean as point of reference), with different components for variance between measurements (within siblings), between siblings (within families), and between families. Random and fixed effects were evaluated with *t* tests and Wald *Z* tests provided by SPSS, at a significance level of 5%. Parameter estimates obtained with MLwiN differed only slightly, and MLwiN's chi-square tests were consistent with the SPSS results.

For each model, we checked whether the longitudinal covariance structure was best described by compound symmetry or by an autoregressive structure, which was decided upon by means of Akaike's information criterion. We also checked whether the fixed regression coefficients should perhaps be considered random and whether first-order interaction effects should be added to the model. These checks involved a large number (10 + 45) of additional significance tests per outcome variable. To prevent too many findings by chance, these tests were carried out at a Bonferroni adjusted level of significance, with which none of the tests were significant.

Finally, percentages of explained variance were calculated for the time effects only, for the confounder variables only, for the coping variables only, and for family functioning only.

Results

Sibling Psychosocial Functioning Compared With Reference Data

Sibling anxiety, QoL, and sibling- and parent-reported behavioral-emotional problems measured with the STAI-C, DuCATQoL, YSR, and CBCL, respectively, could be compared with available reference data of children the same age and sex in the general population. For this purpose, standardized mean differences d (with zero mean and unity variance) were calculated and tested through Student t tests. One month after diagnosis, siblings' anxiety was comparable to that of children the same age and gender ($d = .13, t = 0.90, ns$). Anxiety was lower than the reference group at 6 months ($d = -.40, t = -2.43, p = .02$) and remained low on subsequent measurement occasions (M3: $d = -.39, t = -2.30, p = .03$; M4: $d = -.49, t = -3.08, p = .00$). One month after diagnosis, sibling QoL was significantly lower compared with the reference group ($d = -.59, t = -4.61, p = .00$). Somewhat higher QoL was reported at subsequent measurement occasions, but it remained low compared with the reference group (M2: $d = -.42, t = -2.50, p = .02$; M3: $d = -.32, t = -2.32, p = .02$) until it reached a normal level at 2 years after the diagnosis ($d = -.04, t = -0.30, ns$). Sibling self-reported behavioral-emotional problems assessed with the YSR were higher than the reference group at 1 month, but not significantly ($d = .36, t = 1.58, ns$). Problem scores on the YSR were lower at subsequent measurement occasions (M2: $d = -.12, t = -0.45, ns$; M3: $d = -.39, t = -2.08, p = .047$; M4: $d = -.29, t = -1.75, ns$). When the internalizing component of these problems was examined, it became clear that internalizing problems were relatively high compared with the reference data of the YSR shortly after diagnosis ($d = .51, t = 2.28, p = .03$) but diminished to a normal level during further follow-up. Parent-reported behavioral and emotional problems assessed with the CBCL were comparable to those of the reference group mean at 1 month and were well below the norm at all subsequent measurement occasions (M2: $d = -.20, t = -1.28, ns$; M3: $d = -.41, t = -3.83, p = .00$; M4: $d = -.39, t = -2.63, p = .01$).

Predictors of Sibling Psychosocial Functioning

Fixed effects of the longitudinal mixed-models analyses of sibling psychosocial functioning for confounder and predictor variables are reported in Table II. Deviations from the overall mean at M1, M2, M3, and M4 are displayed in standardized estimates in Figures 2a and 2b.

Anxiety

As assessed by the STAI-C, self-reported anxiety was associated with age, gender, the number of hospital days, predictive and vicarious control, and family adaptation and cohesion. Girls reported relatively more anxiety than boys, and older siblings reported more anxiety than younger siblings. Siblings reported less anxiety as the ill child experienced more hospital days. As to the cognitive coping strategies, it appeared that siblings were less anxious as they were more optimistic (predictive control). Siblings who relied more on the expertise of the medical specialist (vicarious control) reported somewhat higher levels of anxiety. As to the family variables, siblings reported higher anxiety scores as family adaptation and cohesion were higher. The amount of explained variance by the fixed effects in the longitudinal model was 35% (Table II).

Quality of Life

Sibling scores on the DuCATQoL were associated with age, family adaptation, and parent psychological distress. The older they were, the lower siblings' QoL was. Sibling QoL was negatively related to family adaptation and was positively related to parental psychological distress. There were no associations of QoL with cognitive coping. The longitudinal model explained 27.6% of the variance in sibling QoL.

Self-Reported Behavioral-Emotional Problems

Sibling self-reported behavioral-emotional problems on YSR were associated with the fatality of the patient's illness and with family adaptation. Siblings whose ill brother or sister died had reported more psychosocial problems on measurement occasions before the ill child's death. In families with high adaptation levels, siblings tended to report more overall psychosocial problems. The amount of variance explained by the investigated longitudinal model was 40.7%.

Parent-Reported Behavioral-Emotional Problems

Behavioral-emotional problems in siblings as reported by parents on the CBCI, were associated with parental distress only. The more symptoms of distress parents reported, the more psychosocial problems they reported in their well child. The amount of explained variance was low (3.2%).

Insecurity

Age; sex; predictive, vicarious, and interpretative control; and family adaptation and cohesion were all associated with the amount of insecurity experienced on different occasions of measurement with the SSERQ-s. Older

Table II. Parameter Estimates for Multilevel Models of Sibling Psychosocial Functioning Predicted by Measurement Occasion, Sibling and Illness Characteristics, Coping, and Family Functioning, *M* (*SD*)

	Anxiety (STAI-C)	QoL (DuCA TQoL)	YSR total (Self-Report CBCL)	PRF Total (Parent Report CBCL)	Insecurity (SSERQ-s)	Loneliness (SSERQ-s)	Emotional Involvement (SSERQ-s)	Positive Emotions (SSERQ-s)
<i>Fixed effects</i>								
<i>Measurement interval,</i>								
<i>no. of siblings</i>								
At 1 month (M1), <i>N</i> = 83	-.22 (.18)	.19 (.20)	-.45 (.23)	.11 (.26)	-.06 (.17)	-.31 (.18)	-.09 (.18)	.01 (.21)
At 6 months, <i>N</i> = 66	-.47* (.19)	.48* (.20)	-.68** (.23)	.07 (.26)	-.18(.18)	-.30 (.19)	-.20 (.19)	.18 (.22)
At 12 months, <i>N</i> = 60	-.49* (.19)	.43* (.21)	-.77** (.24)	-.14 (.27)	-.14(.18)	-.27 (.19)	-.18 (.19)	.10 (.22)
At 24 months, <i>N</i> = 57	-.58** (.21)	.67** (.22)	-.77** (.24)	-.04 (.27)	-.38* (.19)	-.50* (.20)	-.35 (.20)	.05 (.23)
<i>Sibling^a and illness^b</i>								
Female gender	.52** (.16)	-.35 (.18)	.45 (.23)	-.11 (.23)	.34* (.15)	.40* (.17)	.28 (.16)	.04 (.20)
Age at M1	.19* (.08)	-.43** (.09)	.24 (.14)	.08 (.12)	.15* (.07)	.18* (.08)	.20* (.08)	-.08 (.10)
Diagnosis of leukemia/ lymphoma	.17 (.19)	-.27 (.22)	.49 (.26)	.15 (.30)	-.09 (.19)	.22 (.18)	.06 (.20)	-.19 (.23)
No. hospital days	-.13 (.07)	.01 (.06)	-.02 (.06)	-.03 (.04)	.06 (.05)	.11 (.06)	.05 (.06)	-.03 (.06)
Ill sibling deceased during study	.42 (.27)	-.47 (.31)	.68* (.33)	.12 (.39)	.49 (.27)	.04 (.28)	.21 (.28)	.46 (.33)
<i>Coping</i>								
Predictive control	-.20** (.06)	.10 (.06)	-.06 (.06)	-.01 (.05)	-.37** (.06)	-.19** (.07)	-.16** (.06)	.02 (.07)
Vicarious control	.14* (.07)	-.02 (.07)	.01 (.08)	.03 (.06)	.23** (.06)	.14 (.07)	.18** (.07)	.08 (.07)
Interpretative control	.04 (.06)	-.06 (.06)	-.02 (.07)	-.01 (.05)	.11* (.05)	.11 (.07)	.21** (.06)	.11 (.06)
<i>Family functioning</i>								
Adaptation	.41** (.06)	-.29** (.06)	.21** (.07)	.01 (.05)	.20** (.06)	.30** (.07)	.14* (.06)	.09 (.07)
Cohesion	.13* (.06)	-.10 (.06)	.04 (.08)	.00 (.05)	.11* (.06)	.03 (.07)	.24** (.06)	.12 (.07)
Parent mental health	.01 (.07)	.18** (.07)	.05 (.07)	.14** (.05)	.01(.06)	.02 (.07)	.02 (.07)	.08 (.07)
Percentages of explained variances by fixed effects (full model)	34.7	27.6	40.7	3.2	42.5	23.8	34.9	9.1
Total number of observations in analysis ^c	230	232	135	245	252	252	250	252

STAI = State-Trait Anxiety Inventory for Children; QoL = quality of life; DuCATQoL = The Dutch Children's AZL/TNO Quality of Life questionnaire; YSR = Youth Self-Report; PRF = parent report form; CBCL = Child Behavior Checklist; SSERQ-s = Situation-Specific Emotional Reactions Questionnaire for siblings.

Fixed and random effects are tested for significance with *t* tests; variance and covariance parameters are tested with *t* tests and Wald's *Z* tests, respectively (Rasbash et al., 2000).

p* < 0.05, *p* < 0.01.

^aGender: 0 = boy, 1 = girl.

^bDiagnosis: 0 = solid or brain tumor, 1 = leukemia or lymphoma.

^cThe total number of observations varies from sum of *N* numbers because of missing values.

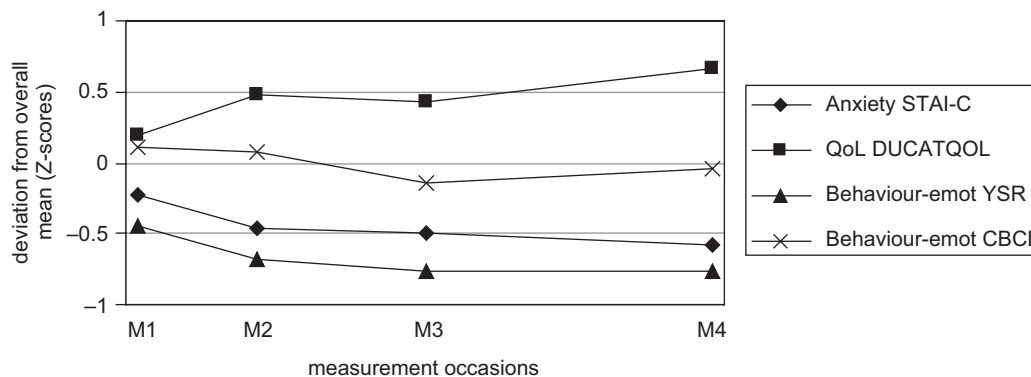


Figure 2a. Estimates of deviations from overall mean over measurement occasions (Z scores): STAI-C, DuCATQoL, YSR, CBCL.

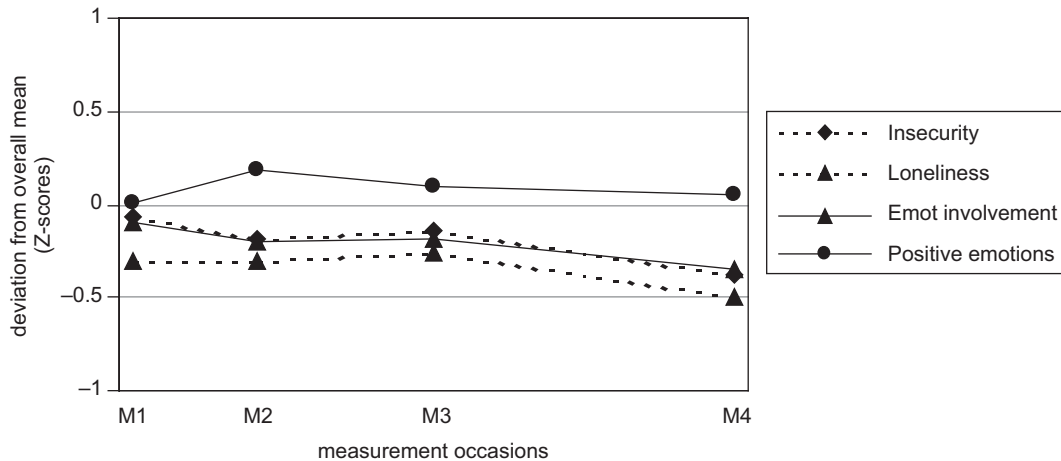


Figure 2b. Estimates of deviations from overall mean over measurement occasions (Z scores): SSERQ-s

siblings and girls reported relatively more feelings of insecurity than younger siblings or boys. As siblings relied more on positive expectations regarding the illness (predictive control), they reported lower levels of insecurity. But siblings who relied more on the medical specialist (vicarious control) and who showed greater efforts to understand the illness (interpretative control) reported more feelings of insecurity. Besides, siblings tended to feel more insecure as higher levels of family adaptation and cohesion were reported. The amount of explained variance within this longitudinal model was 42.5%.

Loneliness

Loneliness experienced by the siblings on the SSERQ-s was associated with sibling age, gender, predictive control, and amount of family adaptability. Older siblings and girls reported more feelings of loneliness compared with younger siblings and boys. Siblings experienced less feelings of loneliness as they remained optimistic about the illness (predictive control). More prominent feelings of loneliness were reported in families with higher levels of adaptation. The amount of explained variance was 23.8%.

Emotional Involvement

Emotional involvement in the illness process as reported by the sibling on the SSERQ-s was associated with age; predictive, vicarious, and interpretative control; family adaptation, and family cohesion. Older siblings reported being more emotionally involved with the illness. As siblings relied less on a positive outcome of the disease (predictive control) they were less likely to be emotionally involved in the illness process, as reported by the sibling. Siblings who tended to rely more on the medical specialist's capability (vicarious control) and who put more effort into trying to understand the illness (interpretative control) were more

emotionally involved in the illness process. Siblings in cohesive families and in families with high adaptation levels reported feeling more emotionally involved as well. The amount of explained variance was 34.9%.

Positive Feelings

The amount of positive feelings experienced by the siblings (SSERQ-s) was associated with none of the investigated variables. The amount of explained variance in this longitudinal model therefore was low (9.1%).

Discussion

The results of the present study show that siblings of pediatric cancer patients, as a group, were most distressed shortly after the diagnosis. In these first weeks, overall QoL was significantly impaired, as was the siblings' emotional well-being. These primary adjustment difficulties diminished rapidly within the first 6 months following the diagnosis in the ill child. The decrease was of such magnitude that means became even lower when compared with reference data, especially for sibling self-reported anxiety and parent-reported behavioral-emotional problems. The general trend of stabilization indicates that siblings show a remarkable resilience in their adjustment to the illness over time. These findings, however, represent group means. Other studies have demonstrated the importance of determining subgroups of siblings with serious adjustment problems instead of focusing on the whole group. Alderfer et al. (2003) found that 30% of siblings of survivors of childhood cancer still had significant symptoms of posttraumatic stress disorder (Alderfer et al., 2003). In a sample of Dutch siblings, of whom the majority were the participants in the present study, 47% of school-age siblings

still reported low overall QoL and 26% of teenage siblings still reported clinically relevant emotional problems 2 years after the ill child's diagnosis (Houtzager, Grootenhuis, Caron, & Last, in press). The determination of children considered at risk in these studies demonstrates that despite overall resilience, a relatively large subgroup of siblings—one third—experiences long-term adjustment problems. Nevertheless, little is known about the characteristics of these at-risk children. The present longitudinal study was conducted in order to investigate how to possibly recognize vulnerable children in terms of individual characteristics such as age, gender, illness specificity, and coping strategies, as well as systemic characteristics such as family cohesion/adaptability and parent mental health.

Firstly, differences in psychosocial adjustment to the illness were found according to sibling age and gender. Older siblings and girls seemed to be most at risk for adjustment problems. Older siblings reported higher levels of anxiety, lower QoL, and more insecurity and loneliness, but also more emotional involvement with the illness. Sisters reported higher levels of anxiety, insecurity, and loneliness than brothers. Girls and older siblings may eventually have more responsibilities, be more involved in the illness, and therefore suffer more from restrictions in their daily lives and overall development. The results indeed show that older siblings were more involved in the illness process, which was related to lower optimism, more reliance on information and the doctor's capabilities, and higher levels of family adaptation and cohesion. Most of these variables were in turn related to more unfavorable adjustment.

The type of diagnosis in the ill brother or sister did not play a significant role in the development of psychosocial adjustment problems in this study group. However, an unfavorable course of the disease and the number of days in the hospital did seem to have their impact. During 2-year longitudinal follow-up of the 56 families that participated, 11 patients died. Results show that teenagers whose ill brother or sister died during the study had reported more overall emotional-behavioral problems on the YSR during the period before the ill child's death. For the other measures of psychosocial adjustment, a trend toward less favorable adjustment in siblings of a deceased patient could be observed. Because of small subgroup sizes, however, no significant associations were found here. The results suggest that siblings are affected not only by the death of the ill child, but also by the unfavorable prognosis and course of the illness per se. Adjustment problems thus manifest themselves at an earlier stage of the illness and need to be

recognized as emotional reactions to the uncertainty of a complicated illness course. The number of days the ill child had to visit the hospital in the previous period seemed to diminish the amount of anxiety that was experienced. Although one could equally argue that the burden of long or frequent hospitalizations would negatively influence siblings' emotional state, the soothing effect of these hospital visits seems to outweigh this burden. Siblings may be comforted by the fact that the ill child is getting actively treated for the illness, relieving their feelings of despair or hopelessness. This may illustrate that as long as something is actively being done to cure the ill child, there is room for hope.

The family system was hypothesized to be an important context in which consequences of the illness would manifest themselves for the siblings of the ill child, as was supported by the present results. Family adaptability was the most prominent family-systems variable affecting sibling adjustment in the present study. Siblings in families with overall high levels of adaptability throughout the disease process showed significantly more adjustment problems on several domains of functioning: anxiety, overall QoL, behavioral-emotional problems, insecurity, and loneliness. This finding is congruent with the basic assumptions of the so-called circumplex model of family functioning developed by Olson, Sprenkle, and Russell (1979), on the basis of which the FACES questionnaire that was used here was developed. In this theoretical framework, *family adaptability* is defined as the tendency of a family system to change its power structure, role relations, and relationship rules in reaction to distress; and *family cohesion* is defined as the amount of closeness and mutual involvement experienced in the family system (Olson et al., 1979). In the circumplex model, moderate levels of cohesion and adaptability are considered to be related to the most favorable adjustment outcome in families faced with stress, whereas extreme levels of adaptation or cohesion are related to less adaptive functioning (Olson et al., 1979). Thus, a balanced family structure that incorporates both stability and change seems to be most functional. Siblings with adjustment problems in the present study came from families with high levels of adaptability, or "chaotic" systems typical of families that have problems competently dealing with stress. Although these findings were congruent with the theoretical framework, they were contrary to findings in cross-sectional studies in which systems variables were related to sibling adjustment. In two studies (Cohen et al., 1994; Horwitz et al., 1990), high adaptability was related to more favorable psychological functioning in siblings. It

has therefore been argued that family structures of extremely high adaptability and cohesion may be more functional in the specific context of childhood cancer (Horwitz et al., 1990). The contradictory results presented here are likely to be due to the longitudinal study design. The two studies in which extreme family adaptability was reported to be favorable were based on cross-sectional data, whereas the present study concerned longitudinal measurement of psychosocial adjustment as well as family functioning. The previous studies indicate that at a certain cross-section in time, adaptability may be a functional systemic coping strategy. However, the present results seem to indicate that the favorable effect of high family adaptability is not present when of longer duration. To be more precise, the family's primary adjustment to the illness may temporarily be realized by changing the family structure, family rules, and role relationships as a function of the illness and of the needs of the ill child. However, it may be harmful for family members should this adaptability become a long-term, chronic state of constantly fluctuating rules and roles, as has been demonstrated here. A long-term chaotic family climate puts a strong claim on all family members. For siblings of a child with cancer, such a family structure requires long-term flexibility, offers no security, and lacks stability and support. This is likely to result in emotional distress. The study results suggest that high levels of emotional involvement in the illness process are associated with high family adaptability, demonstrating the effort that is demanded from siblings in these families. In conclusion, the longitudinal data show that although family adaptability may be effective when transitory, such a process can be harmful for siblings when it becomes structural.

The impact of parental mental health was assessed as well, related to siblings' emotional-behavioral problems as reported by the parent. This finding corresponds with findings in cross-sectional studies (Cohen et al., 1994; Sahler et al., 1994; Sahler et al., 1997). The fact that parental distress was not related to adjustment difficulties reported by the siblings themselves may indicate two different processes. On the one hand, distressed parents are likely to be more sensitive to disruptive or disturbing behavior in the sibling(s) than parents who are less distressed. On the other hand, siblings of distressed parents may be less likely to focus on their own adjustment problems, in order to protect their parents from additional problems to deal with.

In the present study, a model of cognitive control was introduced to investigate the ability of siblings to cope with a relatively uncontrollable event, the diag-

nosis of cancer in a brother or sister. The cognitive coping strategies under investigation predicted sibling adjustment in several ways. Siblings who were able to remain optimistic about the course of the ill child's disease (predictive coping) felt less anxious, insecure, and lonely. This finding stresses the importance of the ability to maintain a positive, optimistic view regarding the illness. The protective power of a positive outlook has been described before (Barbarin, 1988; Koocher & O'Malley, 1981). Positive expectations about the course of the disease may give siblings a sense of mastery or cognitive control. Besides the effect of predictive cognitive coping, interpretative and vicarious cognitive coping played a role in the siblings' feelings of insecurity about the illness. It appeared that siblings who reported high levels of insecurity relied more on interpretative (searching for meaning and understanding) and vicarious (relying on the medical specialist) control. Siblings who feel insecure and anxious may be more likely to have questions regarding the illness and to rely on the specialist's power than would other siblings. On the other hand, the same cognitive coping strategies were related to more emotional involvement with the illness. Siblings who feel more involved may tend to be more interested in the meaning of the illness and have more opportunities to rely on the medical treatment.

There were several limitations to the present study. Firstly, a relatively small sample size (which represents a common problem in this field of research) restricted the number of predictors to be investigated in the longitudinal model and made investigation of interaction effects impossible. The small sample size also restricted the power of this study. As a result, the impact of an unfavorable prognosis that appeared to be a trend could not fully be determined. Besides its small size, the study group suffered from selective dropout. This was due mainly to the fact that 11 patients died during the 2-year follow-up. This resulted in a final study group of siblings of children with more favorable prognoses. The results found here must be interpreted accordingly: Although siblings adjust to the illness relatively well, it must be noted that these are siblings of children who have survived a very serious illness. Another limitation of the study design was the absence of a healthy control group, without a brother or sister with cancer. This limited the interpretability of the results, in terms of specificity of processes for siblings of children with cancer. For example, the interrelation between adjustment and family processes may be specific for siblings of children with cancer or it may be a process that occurs in families in general.

The relationship that was found between optimism and a positive adjustment outcome requires further exploration. The relationship between coping and outcome was investigated on each measurement occasion within the present study design. However, it can be questioned how optimism and a positive emotional state are related. Is optimism really a coping strategy that can be influenced or learned, or is it a traitlike construct that co-occurs with the relative absence of emotional disturbance? Does a positive, optimistic outlook elicit a positive emotional state, or vice versa? Future studies should be conducted to investigate the nature and mechanism of optimism, in order to be able to implement these findings in clinical practice. The role of sibling coping strategies has been investigated only rarely. The use of a sensitive illness-specific instrument that assesses coping separately from emotional outcome is important and has been shown to be useful in the present study. However, the interrelationship of coping with the acuteness of the illness and family dynamics certainly requires further study. The coping strategies that were investigated represented exclusively the siblings' *cognitive* strategies directed at gaining a sense of control over the illness situation. This approach is not at all conclusive, and other strategies of coping, such as seeking social support, distraction, denial, or emotional distancing, were not investigated and deserve to be studied in siblings of a critically ill child. Finally, from a family-systems perspective, the parent's coping may be an important modifier of a sibling's coping resources as well.

The present study's results demonstrate the complexity of family adjustment processes. However, although its longitudinal design was hypothesized to be most suitable to detect processes such as family dynamics, it produced findings that were interpretable but contradictory to those of other studies. Therefore, further assessment of the impact and development of family dynamics over time is recommended. Assessment of changes in the family system as a whole in larger study groups will enable researchers to assess how structures change in families with a seriously ill child during and after the medical treatment, how the roles siblings have in the family change as a result of the illness, how illness variables influence these processes, and how individual family members are affected by these dynamics over time. Comparison with a healthy control group without a child with cancer is needed as well, in order to assess typical high and low levels of adaptability or cohesion in families with a seriously ill child.

The results emphasize that when a child is critically ill, subgroups of siblings are at risk and therefore in need

of extra attention in clinical practice. Firstly, girls and older siblings seem to be at risk for adjustment problems. These may eventually be the siblings with more responsibilities in the illness process. That this can have a negative impact suggests that siblings may lack positive distraction from negative events related to the illness. These siblings may need to be supported and enabled to participate in leisure activities, without feeling guilty, while at the same time being involved in the illness process. Parents need to be informed about how to communicate this coping strategy to their well children.

Secondly, siblings of children with an unfavorable prognosis seem to be at risk. Extra support for these vulnerable children is needed. They and their parents should be closely monitored and guided until well after the death of the ill child. However, further study of specific needs of this subgroup is recommended as well. Thirdly, it appeared that adjustment problems in siblings were embedded in family dynamics. Therefore, interventions need to be focused on the family system as a whole. A certain level of stability and balance in the family system are of utmost importance to the sibling's well-being and ability to adapt to long-term distress and changes related to the illness and treatment. When an unstructured family constellation is diagnosed, the practitioner needs to assess to what extent this is the result of high levels of distress caused by the illness, and whether it is a transitional phase or a structural feature of this particular family. If such adaptability becomes structural, a family therapist may be needed to help the family through this phase or to try to establish a more balanced family climate in which family members can feel safe and secure and each family member's needs can be sufficiently met.

Finally, the results show that siblings need to be offered opportunities to remain optimistic or positive about the ill child's treatment and prognosis. Parents as well as general practitioners, pediatricians, nurses, and other professionals should be adequately informed about this need. They need to be instructed on how to provide siblings with realistic information about the illness, if the situation allows this, without undermining the possibility of a positive outcome of the illness process. Parents may find it difficult to provide children with information about the illness, because this is contrary to their natural need to protect their child. However, the results show that siblings who feel insecure tend to long for information about the illness and may want to be involved in the illness process. If parents keep information from their child, he or she can be convinced that the illness is too threatening to talk about.

This may only cause unrealistic fear regarding the illness and enhance feelings of isolation, guilt, and resentment (Spinetta et al., 1999). It is therefore important that parents be guided in what and how to tell siblings about the illness/treatment process and to involve them in discussions about it. This applies especially to young children. In support groups, vulnerable siblings can learn cognitive and other coping strategies that are directed at maintaining a positive outlook. Despite the realistic fears and feelings of insecurity that result from the illness, siblings can become more familiar with different aspects of the illness in a secure setting, ventilate feelings, and experience support from other siblings their own age. Several studies have demonstrated positive results with support groups for siblings (Carpenter, Sahler, & Davis, 1990; Dolgin, Somer, Zaidel, & Zaizof, 1997; Heiney, Goon-Johnson, Ettinger, & Ettinger, 1990; Houtzager, Grootenhuis, & Last, 2001; Kinrade, 1985; Lobato & Kao, 2002).

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