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Family Functioning Mediates Adaptation in Caregivers of Individuals with Rett Syndrome

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Abstract

Objective—The objective of this study was to investigate factors related to family functioning and adaptation in caregivers of individuals with Rett syndrome (RS).

Methods—A cross-sectional quantitative survey explored the relationships between demographics, parental self-efficacy, coping methods, family functioning and adaptation. A forward-backward, step-wise model selection procedure was used to evaluate variables associated with both family functioning and adaptation. Analyses also explored family functioning as a mediator of the relationship between other variables and adaptation.

Results—Bivariate analyses ($N=400$) revealed that greater parental self-efficacy, a greater proportion of problem-focused coping, and a lesser proportion of emotion-focused coping were associated with more effective family functioning. In addition, these key variables were significantly associated with greater adaptation, as was family functioning, while controlling for confounders. Finally, regression analyses suggest family functioning as a mediator of the relationships between three variables (parental self-efficacy, problem-focused coping, and emotion-focused coping) with adaptation.

Conclusion—This study demonstrates the potentially predictive roles of expectations and coping methods and the mediator role of family functioning in adaptation among caregivers of individuals with RS, a chronic developmental disorder.

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Practice Implications—Practice Implications: A potential target for intervention is strengthening of caregiver competence in the parenting role to enhance caregiver adaptation.

Keywords

adaptation; family functioning; Rett syndrome; coping; self-efficacy; caregiver; neurodevelopmental disorder

1. Introduction

Rett syndrome (RS) is a panethnic neurodevelopmental disorder, and is one of the major causes of severe intellectual disability in females, affecting approximately one in 10,000 [1,2,3]. RS is characterized by the loss of intellectual functioning, as well as fine and gross motor skills (e.g., purposeful hand use, communicative abilities) after a period of seemingly normal development. Individuals with RS can also experience a deceleration of head growth, stereotypic hand movements, seizures, respiratory dysfunction, and ataxic or apraxic gait [2,4]. During the rapid destructive stage of RS, which is usually initiated between one and four years of age, children can experience disturbing behavioral problems that can be very trying for their families including, for example, episodes of screaming, panic attacks, and self-injurious behavior [2,5]. Accordingly, caregivers of individuals with RS can face an array of psychosocial challenges.

It has been shown that parents of girls with RS experience more stress [6] and less satisfaction with their marital relationships [4] than controls. While one study reported normative levels of depression in mothers of girls with RS [6], most evidence suggests an elevated risk for depression and reduced health-related quality of life [7]. Another study showed that better mental health in mothers of girls with RS was associated with better family functioning [8]. Family functioning is a dynamic phenomenon that has been described as a family's ability to achieve goals integral to the lives of its members [9]. While there have been no studies of the relationships between parental self-efficacy, coping strategies, and adaptation in families of girls with RS, some of these associations have been established in studies looking at parents of children with other genetic disorders [10,11].

Thompson's Transactional Stress and Coping model [10,12,13,14], provides a useful framework for understanding how an individual adapts to a stressor within the family. The cognitive processes of appraisals and expectations, methods of coping, and family functioning are hypothesized to mediate adaptation to being a caregiver of an individual with a chronic condition in Thompson's model. There is not sufficient evidence for claiming path relationships among these constructs, but this model suggests a transactional rather than a unidirectional relationship in which each factor impacts the others over time. The objectives of the current study were to (1) investigate potential relationships among parental self-efficacy, coping methods, family functioning, and caregiver adaptation, and (2) evaluate family functioning as a mediator of the relationships between potential predictor variables and caregiver adaptation.

Per the model depicted in Figure 1 [15], we specified the following hypotheses regarding relationships of other predictor variables and family functioning: higher levels of parental

self-efficacy, a greater proportion of problem-focused coping, and a lesser proportion of emotion-focused coping will be associated with more effective family functioning (relationship *A*). Regarding relationships of predictor variables with adaptation, we made the following hypotheses: higher levels of parental self-efficacy, a greater proportion of problem-focused coping, and a lesser proportion of emotion-focused coping will be associated with greater caregiver adaptation (relationship *C*), as will more effective family functioning (relationship *B*). Finally, we hypothesized that family functioning would be a mediator of the relationships between adaptation and both parental self-efficacy and coping methods (relationship *C* vs. relationship *D*).

2. Methods

2.1 Sample

The study population consisted of men and women who self-reported as the primary caregivers of a child with RS, and who were able to read and write in English. Cohabitation with the child with RS was an additional inclusion criterion. To attain a diverse representation of the population, participants were recruited from four clinics across the eastern and southeastern United States, the 25th Annual Education & Awareness Conference for Rett Syndrome, and various RS organizations and websites. All interested individuals were invited to complete the paper surveys that were mailed to their homes or to visit the online version available on a SurveyMonkey® platform. Participants were compensated with a modest gift card. Informed consent was obtained from all participants, and the study was approved by ethics committees from all involved institutions.¹

2.2 Measures

This study used a cross-sectional research design with a quantitative survey that was available in both paper and electronic versions. The survey was pilot tested to determine the clarity and comprehensibility of questions, as well as to gain feedback on content. In addition to demographics questions, the survey instrument was composed of validated measures of parental self-efficacy, coping methods, family functioning, and adaptation.

Demographic Information collected included age, gender, race, ethnicity, marital status, education, income, perceived finance burden (extent of agreement with “Healthcare costs for RS are a financial burden to me and my family”), workload (“How much of the work do you share with other members in your family with regard to caring for you child with RS?” contrasting responses indicating all/most of the work with responses indicating some degree of sharing), health of caregiver (5-point item ranging from “poor” to “excellent”), amount of caregiver sleep per night, relationship to child with RS, number of children with RS, presence of children requiring special services, number and type of adults living in the household, frequency of information-seeking behavior (4-point item ranging from “never” to “regularly”), and use of support programs. The current age of the individual with RS and

¹This study was approved by the National Human Genome Research Institute (NHGRI) Institutional Review Board (IRB; Protocol # T-HG-0021), the Johns Hopkins Bloomberg School of Public Health IRB (Protocol #: IRB00001946), the University of Alabama at Birmingham IRB (Protocol #X090417004), the Self Regional Healthcare IRB (IRB associated with Greenwood Genetic Center), and the Baylor College of Medicine (Protocol #H-14884). Approval was also obtained from the research compliance reviewer at the Kennedy Krieger Institute.

his/her age at diagnosis were also recorded. In addition, two 5-point items were used to assess the caregiver's subjective impression of the severity of their child's condition compared to children of the same age with RS ("not at all severe" to "very severe") and without RS ("doing a lot worse" to "doing a lot better"). This information was used to evaluate these variables' potential as confounders in linear regression analyses, as well as to assess external validity.

Parental Self-Efficacy was assessed using the Efficacy component of the Parenting Sense of Competence Scale (PSOC) [16], which indicates the degree to which the parent feels competent, capable of problem solving, and familiar with parenting. Each of the seven items allowed for responses on a 6-point scale ranging from "strongly disagree" to "strongly agree." This subscale has previously been applied as an independent measure, with a reported internal consistency of 0.71 [17]. Summed scores could range from 7–42, with higher scores indicating greater parental self-efficacy.

Coping Methods were assessed using the Ways of Coping Checklist-Revised (WCC-R) [18]. Respondents were asked to estimate how often they use each of the 42 coping strategies, classified under the following five domains: problem-focused, wishful thinking, seeks social support, blamed self, and avoidance. This scale has been shown to be valid and reliable with Cronbach's alphas of the subscales ranging from 0.73–0.88 [18]. As proposed by Zakowski and colleagues [19], the blamed self, wishful thinking, and avoidance subscales were combined into one emotion-focused coping score. Finally, the two proportions were calculated by dividing the score sums of problem- and emotion-focused coping separately into the total sum of all coping strategies used.

Family Functioning was assessed using the Family Assessment Measure III (FAM-III). The FAM-III is based on the Process Model of Family Functioning and is used to distinguish areas of family strengths and weaknesses, as well as to differentiate families that are coping successfully from those utilizing less effective coping styles [20]. The General Scale contains 50 items measuring seven constructs (task accomplishment, role performance, communication, affective expression, affective involvement, control, and values and norms), and contains two additional response style subscales (social desirability and defensiveness). The FAM-III has been demonstrated to have acceptable internal reliability (Cronbach's $\alpha=0.93$ for the overall rating) [9]. The subscale scores were summed to yield an overall Family Functioning score, which could range from 0–150. Higher scores indicating less effective family function suggest an assessment of family dysfunction. In accordance with the developers' design, "more effective family functioning" will refer to lower scores on this measure.

Adaptation was assessed using the Psychological Adaptation Scale (PAS) [21]. This 20-item measure assesses the individual's adaptation at a particular time, and captures four domains of adaptation: Positive Social Impact, Positive Self-Concept/Esteem/Worth, Positive Stress Response Symptoms, and Positive Spiritual/Existential Impact. Validity data for these four dimensions comes from six studies [21]. Confirmatory factor analysis suggests that each of the four domains supports the single, second-order construct of adaptation [22]. Summed scores could range from 0–5, with higher scores indicating greater adaptation.

2.3 Data Analysis

Data were analyzed using SPSS 16.0. The primary outcome variables, family functioning and adaptation, were analyzed in parallel. As appropriate, Pearson's correlation, t-test, or ANOVA was used to test the associations between each covariate and the outcome variables. Multivariable linear regression modeling was used to test associations between each predictor and the outcome variables: a forward-backward, step-wise model selection procedure was used to determine inclusion of potential confounders ($p < 0.2$). The standardized β coefficients with and without family functioning included in each of the models were compared to assess family functioning as a potential mediator of the relationships between the other predictor variables and adaptation. Finally, PROCESS for SPSS (version 2.15) was used to assess the significance of the mediator's indirect effect by calculating bias-corrected 95% confidence intervals derived from 1000 bootstrap resamples [23]. In all analyses the following demographic variables and confounders were initially tested for inclusion: age, gender, race, ethnicity, marital status, education, income, perceived finance burden, workload, health of caregiver, amount of caregiver sleep, respondent's relationship to child(ren) with RS, number of children with RS, age of child(ren) with RS, number of other children requiring special services, adult cohabitants, frequency of information-seeking behavior, use of support programs, and years asymptomatic.

3. Results

During the recruitment period from May to October 2009, 208 individuals visited the survey website, 104 individuals returned paper surveys at the annual educational conference, and 152 individuals returned paper surveys by mail. There were no statistically significant demographic differences between these three groups. Of the 426 total individuals that began the survey, 26 individuals were excluded from analysis because a majority of the survey sections were not completed or because they did not complete one or both of the outcome measures. These 26 individuals did not differ from the rest of the sample in terms of demographics including age, education, and age of child. Forty-eight additional participants were excluded from regression and mediation analyses due to incomplete data on the predictor, mediator, or outcome variables.

3.1 Sample Characteristics

The average age of participants was 43.28 ± 9.68 years and ranged from 22–74 years ($n=396$). More than half of the caregivers reported seeking information about RS on a regular basis, and the majority of caregivers had utilized at least one support program (75%). Demographic characteristics of caregivers are shown in Table 1.

The vast majority of participants (98.7%) had only one child with RS ($n=392$). Regarding characterization of the individuals with RS, more than 98% were females, and the average age was 12.6 ± 9.4 years (ranging from 1.6–50 years). The average age of diagnosis for the child with RS was 4.2 ± 4.1 years (ranging from 0.9–30 years), whereas the average age that symptoms were first noticed was 1.4 ± 1.1 years (ranging from 0–12 years). When compared to other children of the same age with RS, caregivers reported that their child with RS had a somewhat less severe presentation, with a mean perceived severity score of 2.74 ± 1.05 out

of a possible 5 points. Means and standard deviations for the three key predictor variables and the two outcome variables are shown in Table 2.

3.2 Variables associated with Family Functioning (relationship A)

To evaluate the relationships between the hypothesized predictor variables and family functioning, bivariate analyses were performed and demonstrated that those caregivers who did the majority of the work with regard to the care of the child with RS had less effective family functioning than those caregivers who shared more of the caregiving responsibilities with other family members ($t=4.789$, $p<0.001$). Less effective family functioning was also associated with having more than one child with RS ($r=0.11$, $p=0.026$) and with having children who were older when symptoms of RS emerged (i.e., more years asymptomatic; $r=0.12$, $p=0.021$). Finally, it was also found that three key variables were significantly correlated with family functioning: parental self-efficacy ($r=-0.225$, $p<0.001$), problem-focused coping ($r=-0.296$, $p<0.001$), and emotion-focused coping ($r=0.417$, $p<0.001$).

These relationships were then tested individually in a series of linear regressions while controlling for all statistically significant confounders ($p \leq 0.2$), which yielded the models summarized in Table 3. The proportion of emotion-focused coping accounted for an additional 11.2% of the variance when added to the model, the problem-focused coping proportion accounted for 5.3%, and parental self-efficacy accounted for 3.2%.

3.3 Variables associated with Adaptation (relationships B & C)

To evaluate the relationships between the hypothesized predictor variables and adaptation, bivariate analyses were performed and demonstrated that those caregivers who were the biological mother had significantly higher levels of adaptation than other respondents ($t=2.515$, $p=0.015$). Greater adaptation was also associated with having non-parental, adult cohabitants ($t=-3.177$, $p=0.002$) and with identifying as non-white ($t=-3.332$, $p=0.002$). Finally, it was found that four key variables were significantly correlated with adaptation: parental self-efficacy ($r=0.377$, $p<0.001$), problem-focused coping ($r=0.215$, $p<0.001$), emotion-focused coping ($r=-0.289$, $p<0.001$), and family functioning ($r=-0.262$, $p<0.001$).

These relationships were then tested individually in a series of linear regressions while controlling for all statistically significant confounders ($p \leq 0.2$), which yielded the models summarized in Table 4. Parental self-efficacy accounted for an additional 12.1% of the variance when added to the model, the emotion-focused coping proportion accounted for 9.2%, family functioning accounted for 7.2%, and the problem-focused coping proportion accounted for 5.3%.

3.4 Mediation (relationships C vs. D)

Using a series of linear regressions with each of the key variables along with the potential confounders, it was shown that parental self-efficacy, problem-focused coping proportion, and emotion-focused coping proportion were all significantly associated with adaptation (column C – Table 5). Once family functioning was added to the model, the magnitude of each of the β coefficients became smaller (column D – Table 5). Finally, as the bootstrap confidence interval for each of the indirect effects does not cross zero, our data provides

evidence for our hypothesis that family functioning is a significant partial mediator of the relationships between adaptation and three key variables: parental self-efficacy, problem-focused coping, and emotion-focused coping.

4. Discussion & Conclusion

4.1 Discussion

The findings provide evidence that family functioning partially mediates the relationships between adaptation and three variables: parental self-efficacy, problem-focused coping, and emotion-focused coping. Thompson and colleagues found that family functioning is related to adjustment in mothers of children with sickle cell disease [14]. Family functioning has also been found to mediate the relationship between benefit-finding and depressive symptoms among mothers of children with an autism spectrum disorder [24]. However, this is the first study that has demonstrated the mediator role of family functioning with a theoretically based, multiple-domain measure of adaptation such as the PAS. Our findings suggest that parental self-efficacy, problem-focused coping, and emotion-focused coping not only have a direct relationship with caregiver adaptation, but are also associated with adaptation through their relationship with family functioning. However, because this study was cross-sectional, inferences cannot be made about how family functioning and adaptation change over time, nor about how the relationships between these variables change with the passage of time. Given the findings presented here, a longitudinal investigation is warranted. In addition, future research using qualitative interviews or mixed methods could provide additional insights into the observed relationships.

The important relationships among these variables could be further investigated by applying the step-wise regression analyses utilized here with an *a priori* hierarchical order reflecting theoretically based causal hypotheses to a distinct sample of caregivers. Cross-validation with caregivers of individuals with a neurodevelopmental disorder that presents challenges comparable to those of RS would allow for more substantive conclusions.

We have evidence that our sample was representative in terms of the primary appraisals, the expectations, and the outcome variables assessed at the time point at which the surveys were completed. Means for these variables (Table 2) and demographics (Table 1) were comparable to the data available for this population. There is normative data on self-efficacy among parents of girls aged four to six, which shows average scores of 25.08 ($SD=5.97$) for mothers and 25.77 ($SD=5.29$) for fathers [17]. Our mean of 30.54 ($SD=5.93$) is comparable to these population standards, though it's trending toward greater than normal parental self-efficacy. The mean family functioning score in this study was 50.59 (95% CI: 49.71, 51.46). According to the FAM-III manual, the majority of scores should fall between 40 and 60 [25]. Finally, the mean adaptation score of 3.80 (95% CI: 3.73, 3.88) indicated levels of adaptation similar to those previously reported in comparable populations. Nevertheless, it is possible that parents with higher levels of distress may have been more motivated to complete the survey, or that parents with lower levels of distress may have been more likely to participate due to greater cognitive and emotional availability. While we recognize the potential for selection bias, there is insufficient evidence to categorize the direction.

Since the majority of participants in this study were white, married, well-educated biological mothers of the child with RS, there is a need to study a more diverse sample. Also, it could be informative to capture perspectives of other family members or extra-familial objective assessors, as this study was only able to capture the primary caregiver's evaluation. Fathers are often under-represented in caregiver studies, and the fathers in the current study are likely not representative of all fathers of children with RS, as our target demographic was *primary* caregivers. It would be of interest to capture both parents' perspectives. The actor-partner interdependence model could be used to frame studies to assess the influence of one parent's efficacy and coping on another member's perception of family function and adaptation, and has been used to assess mediation in dyadic datasets [26]. Given the ongoing call for health care professionals to focus not solely on the patient, but on the family as a whole, it could be beneficial to incorporate multiple perspectives into development of comprehensive care guidelines.

Given that many studies have shown that problem-focused coping is more adaptive [10,27], it was somewhat surprising to find that the vast majority of caregivers in this population (over 90%) reported more frequent utilization of emotion-focused coping. Studies of caregivers of children with autism spectrum disorders and of caregivers of adults with Parkinson's disease or dementia have also found a preponderance of emotion-focused coping [28,29,30]. Potential explanations for this include the "goodness-of-fit" hypothesis [19]. According to this reasoning, if the stressor is appraised as changeable or controllable, problem-focused coping is considered more appropriate; however, if the stressor is appraised as unchangeable or uncontrollable, emotion-focused coping is more effective. The fact that caregivers in the current study are using more emotion-focused coping strategies may suggest that they are appraising many of the stressors of being a caregiver as unchangeable or uncontrollable. Nevertheless, emotion-focused coping was inversely associated with both family functioning and adaptation, which is consistent with the literature [13].

Three confounders emerged as significant factors related to family functioning: caregivers who shared more of the workload with regard to caring for their child, who only had one child with RS, and whose child showed symptoms of RS earlier in life tended to have more effective family functioning. Regarding the last confounder, a study of Taiwanese families with Duchenne muscular dystrophy reported a similar relationship [31]. It's possible that being confronted with the challenges of being a caregiver before having time to establish family dynamics ameliorates the difficulty in reorganizing roles and responsibilities when the child starts showing symptoms. In addition, three significant confounders were related to adaptation including relationship to child, presence of non-parental adults in the household, and race of caregiver. Other studies have consistently found that mothers have lower levels of well-being and higher levels of distress than fathers [32,33,34,35]. One potential explanation for the surprising finding that biological mothers in our study tended to have higher levels of adaptation is that the mothers in the current study assumed more traditional parental roles, and therefore were less likely to report inability to adapt because it could have been viewed as a reflection on themselves as parents. It is also possible that this finding says more about the nature of the participating fathers. Because only parents with the most contact with the child were asked to participate, it is possible that the fathers in this study were more engaged than the typical father. In a small study of parents of infants with liver

disease, fathers who reported being highly engaged with the child expressed more anxiety about the child than did mothers [36]. Having more adults in the household to share the responsibilities of taking care of a child with RS may help the primary caregiver to adapt more successfully, possibly explaining that finding. Being non-white was also related to greater adaptation. Similar findings have been reported in studies comparing Latina with non-Latina caregivers and comparing white with African American caregivers [37,38]. Other cultures may foster more interdependence among family members, and therefore the dependency of the child with RS may be less troublesome for these caregivers compared to white caregivers.

4.2 Conclusion

This cross-sectional study of caregivers of children with RS explored the relationships between parental self-efficacy, coping methods, family functioning and adaptation. Parental self-efficacy, the proportions of problem-focused coping and of emotion-focused coping used by caregivers were found to be associated with family functioning. Parental self-efficacy, problem-focused coping, emotion-focused coping, and family functioning were found to be associated with caregiver adaptation. Finally, family functioning was found to be a partial mediator of the relationships between adaptation and three key variables: parental self-efficacy, problem-focused coping, and emotion-focused coping. Although future studies are needed, interventions focused on increasing parental self-efficacy, employing appropriate coping strategies, and facilitating greater family functioning may enhance caregiver adaptation.

4.3 Practice Implications

Because these key variables are theorized to be antecedents of adaptation, tailored interventions aimed at increasing use of appropriate coping, enhancing parental self-efficacy, and helping caregivers' families function more effectively may help caregivers adapt to their child's condition. Health care providers can refer to therapies such as parent management training (PMT) which helps parents become more actively involved in interventions that strengthen family functioning and has been shown to increase parental self-efficacy in a study of parents of children with Asperger syndrome [39]. One study showed that reductions in stress and increases in coping self-efficacy can be achieved in an HIV+ population through a coping effectiveness training (CET) intervention teaching individuals how to identify emotion-focused coping strategies (e.g., cognitive reframing, relaxation) and problem-focused coping strategies (e.g., problem-solving, communication skills), and how to "fit" stressful situations with the appropriate coping methods [40]. Our results indicate that enhancing family functioning through education about and promotion of healthy coping could have profound effects on a caregiver's ability to adapt. Some important roles for the health care provider beyond informing family members about the condition and relevant resources could be (1) to educate about the importance of family dynamics, (2) to facilitate effective communication about roles and responsibilities, and (3) to encourage other members of the family to provide support for the primary caregiver. Health care providers, including genetic counselors, can target each of these areas as well as provide referrals to support groups and other resources to promote adaptation.

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Highlights

- Self-efficacy, problem-, and emotion-focused coping predict family functioning.
- Self-efficacy, coping methods, and family functioning predict adaptation.
- Family functioning mediates the relationship between self-efficacy and adaptation.
- It also mediates the relationship between emotion-focused coping and adaptation.

Figure 1.

This mediation model was adapted from Gelfand and colleagues [16]. Predictor variables include parental self-efficacy, problem-focused coping, and emotion-focused coping. *C* denotes the relationships between predictor variables and adaptation without family functioning included in the model, whereas *D* denotes the relationship including family functioning in the model.

Table 1

The demographic characteristics presented here are those that were significant confounders controlled for in regression analyses.

Demographic Characteristics of Caregivers

Characteristic	Response Option	<i>n</i>	%
Race (<i>n</i> =391) *	White	364	93.1
	Non-White	31	7.9
Ethnicity (<i>n</i> =388)	Not Hispanic or Latino	364	93.8
	Hispanic or Latino	24	6.2
Marital Status (<i>n</i> =398)	Married	339	85.2
	Not Married	59	14.8
Highest Level of Education (<i>n</i> =397)	High School / GED	49	12.3
	Some College	98	24.7
	Bachelor's Degree	134	33.8
	Post-Graduate Degree	94	23.7
	Other	22	5.5
Income (<i>n</i> =390)	Under \$30,000	42	10.8
	\$30,001–\$50,000	60	15.4
	\$50,001–\$70,000	71	18.2
	Above \$70,000	217	55.6
Perceived Finance Burden (<i>n</i> =394)	Strongly disagree	42	10.7
	Disagree	74	18.8
	Neither agree nor disagree	128	32.5
	Agree	103	26.1
	Strongly agree	47	11.9
Workload (<i>n</i> =398)	I do all of the work	58	14.6
	I do most of the work	220	55.3
	Complete sharing of the work	104	26.1
	I do some of the work	15	3.8
	Other family members do all of the work	1	0.3
Health of Caregiver (<i>n</i> =396)	Excellent	32	8.1
	Very good	127	32.1
	Good	160	40.4
	Fair	70	17.7
	Poor	7	1.8
Amount of Caregiver Sleep (<i>n</i> =397)	> 8 hours	11	2.8
	6–8 hours	294	74.1
	3–5 hours	90	22.7

Demographic Characteristics of Caregivers

Characteristic	Response Option	<i>n</i>	%
	< 3 hours	2	0.5
Relationship to Child (<i>n</i> =398)	Biological Mother	352	88.4
	Adoptive Mother	9	2.3
	Biological Father	28	7.0
	Adoptive Father	3	0.8
	Other	6	1.5
Children with RS (<i>n</i> =397)	One child with RS	392	98.7
	More than one with RS	5	1.3
Special Services (<i>n</i> =396)	Other children requiring special services	42	10.6
	No other children requiring special services	354	89.4
Household Adults (<i>n</i> =398)	Parents Only	299	75.1
	Other	99	24.9

Other demographics collected, but not presented in this table, include age of child(ren) with RS, years asymptomatic, information-seeking behavior, and use of support programs. Percentages may not equal 100 (denoted by an *), as participants were allowed to choose more than one response for some items.

Table 2

Each of the means for these five constructs was similar to the normative values obtained from our review of relevant literature.

Distributions for Predictor and Outcome Variables

Construct	possible scores	mean	SD	n
Parental Efficacy	7–42	30.54	5.93	388
Problem-focused coping proportion	0–100%	26.32%	6.16%	390
Emotion-focused coping proportion	0–100%	51.47%	10.24%	390
Family functioning	0–150	50.59	8.82	391
Adaptation	0–5	3.804	0.78	388

Table 3

Variables controlled for: income, relationship to child, number of children with special services, workload, health of caregiver, and perceived finance burden. The R-squared from the regression model of confounders alone was 0.162. “Added” R² refers to the additional variance explained by including the given variable, and was calculated as the difference between the models including and excluding said variable.

Linear Regression Models of Each Key Variable on Family Functioning

VARIABLE	B (SE)	β	p-value	Model R ²	“Added” R ²
Consequences on life of individual with RS	-0.03 (0.90)	-0.002	0.974	0.163	0.001
Consequences on life of caregiver	1.06 (0.77)	0.07	0.170	0.167	0.005
Parental Efficacy	-0.27 (0.08)	-0.18	<0.001	0.194	0.032
Problem-focused Coping Proportion	-34.33 (6.98)	-0.24	<0.001	0.215	0.053
Emotion-focused Coping Proportion	29.77 (4.05)	0.35	<0.001	0.274	0.112

Table 4

Variables controlled for: race, ethnicity, adult cohabitants, relationship to child, years asymptomatic, and amount of caregiver sleep. The R-squared from the regression model of confounders alone was 0.104. “Added” R² refers to the additional variance explained by including the given variable, and was calculated as the difference between the models including and excluding said variable.

<i>Linear Regression Models of Each Key Variable on Adaptation</i>					
VARIABLE	B (SE)	β	p-value	Model R ²	“Added” R ²
Consequences on life of individual with RS	−0.04 (0.07)	−0.03	0.596	0.105	0.001
Consequences on life of caregiver	−0.13 (0.08)	−0.08	0.104	0.113	0.009
Parental Efficacy	0.05 (0.01)	0.36	<0.001	0.225	0.121
Problem-focused Coping Proportion	3.04 (0.64)	0.24	<0.001	0.157	0.053
Emotion-focused Coping Proportion	−2.38 (0.37)	−0.31	<0.001	0.196	0.092
Family Functioning	−0.03 (0.01)	−0.28	<0.001	0.176	0.072

Table 5

Variables controlled for included race, ethnicity, adult cohabitants, relationship to child, years asymptomatic, and amount of caregiver sleep. All β coefficients were significant at $p < 0.001$.

Linear Regressions of Adaptation on Key Variables

Variable	Standardized β Coefficients		Indirect Effects (95% CI)
	C (Family Functioning <i>not</i> included)	D (Family Functioning included)	
Parental Efficacy	0.36	0.32	0.04 (0.01, 0.07)
Problem-focused Coping Proportion	0.24	0.17	0.07 (0.03, 0.11)
Emotion-focused Coping Proportion	-0.31	-0.24	-0.07 (-0.12, -0.03)