

# Caring for the Caregivers: An Investigation of Factors Related to Well-Being Among Parents Caring for a Child with Smith-Magenis Syndrome

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**Abstract** Smith-Magenis syndrome (SMS) is a complex disorder characterized by numerous challenges, including intellectual disability, speech delay, decreased pain sensitivity, sleep disturbances, hyperactivity, mood instability, and self-injury. Caregivers must readily adapt to the ever-changing needs of the child. Due to these demands, caregivers may encounter difficulties maintaining their own level of well-being. Thus, a total of 112 primary caregivers (i.e., parents) of individuals diagnosed with SMS responded to online questionnaires to assess demographic and psychosocial factors, such as perceptions of child health vulnerability, benefit finding, sleep behaviors, anxiety and depression symptomatology, and caregiver satisfaction and self-efficacy, which may be related to caregiver well-being. Results show that, among mothers, caregiver well-being was directly related to perceived child health vulnerability, caregiver satisfaction, and benefit finding, and a significant moderating effect was observed for depression/anxiety counseling after beginning the caregiver role on the relationship between anxiety symptomatology and caregiver well-being. Results further

suggest that maternal caregivers who report high levels of anxiety but do not seek counseling fair the worst in terms of well-being. Among fathers, lower depression symptoms and greater benefit finding were related to higher levels of caregiver well-being. These data show that many factors play roles in influencing coping and well-being among SMS caregivers. Investigating these variables and relationships may reveal additional resources and interventions to assist primary caregivers.

**Keywords** Primary caregiver · Benefit finding · Caregiver well-being · Smith-Magenis syndrome · Depression · Anxiety · Sleep disturbance

## Introduction

“Caring for my child with SMS [is the] most challenging experience thus far in life. I have seen aspects of myself that I’ve hated and other [aspects have] surprised me. [I’ve] felt anger and frustration... but I’ve also experienced love and compassion that is immeasurable.”

- Mother of 21-year-old diagnosed with SMS

Smith-Magenis syndrome (SMS) is a chromosomal disorder caused by a deletion of chromosome 17p11.2 or a mutation of the *RAI1* gene, which lies within the SMS critical region (Elsea and Girirajan 2008; Smith et al. 2006). This syndrome affects approximately 1:25,000 births worldwide and is typically a sporadic syndrome not inherited from a parent. Diagnosis is made by chromosome or DNA analysis (Elsea and Girirajan 2008). Common features among those diagnosed with SMS include cranio-

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facial anomalies, a hoarse voice, feeding problems in infancy, hypotonia, developmental delays, mental retardation, early speech delays, decreased sensitivity to pain, chronic sleep disturbances, hyperactivity, arm hugging, hand squeezing, attention problems, sudden mood changes, and self-injurious behaviors (Elsea and Girirajan 2008; Smith et al. 2006).

Caring for a child with SMS requires that the caregiver be able to readily adapt to these physical and behavioral features as they fit within the typical developmental experiences all individuals face across the lifespan. Particularly difficult are the significant chronic sleep disturbances, the self-injurious behaviors, and explosive temper tantrums. Not only can the caregiver role be challenging on a daily basis, it is a role that the caregiver often assumes for a lifetime given that individuals with SMS live well into adulthood and are usually unable to care for themselves independently. Based upon these facts, and as expressed in the preceding quote, caregivers of children diagnosed with SMS likely encounter challenges maintaining their own level of well-being, including potential challenges with depression, anxiety, and overall well-being.

Little is known about family dynamics and caregiver adaptation among families coping with a SMS diagnosis. To date, a limited number of studies have shown that families of children with SMS and other genetic disorders experience a broad range of challenges in caring for their child (Dyson 1991, 1993; Fidler et al. 2000; Fidler and Hodapp 1998; Hodapp et al. 1998; van den Borne et al. 2006). This research showed that these families are more likely to experience parent-child relational problems than families coping with Down syndrome or Williams syndrome. Parents caring for a child with SMS also report higher levels of pessimism than those caring for children with other genetic disorders. In 2000, Fidler and colleagues reported that some of this increased family stress may be attributed to the heightened number of maladaptive behaviors (e.g., self-injury, sleep disturbance) displayed in individuals with SMS as compared to other children with genetic disorders (Fidler et al. 2000). Similar results were produced several years earlier with research suggesting that stress in families of children with SMS was much higher than families of children with developmental delay of mixed and unknown etiologies (Dyson 1991, 1993). However, previous studies of individuals with chronic illnesses have shown that individuals who perceived more benefits from their illness actually suffered less (Katz et al. 2001); thus, an exploration into benefit finding among primary caregivers for individuals with a lifelong condition may have similar experiences.

Regardless of the ups and downs of childrearing, one of the most important aspects of caregiving is taking care of oneself. Each year in the United States, thousands of

families cope with the many caregiver responsibilities associated with raising a child diagnosed with SMS (Smith et al. 2006). Despite initial findings, researchers have not formally investigated key aspects of caregiver well-being and salient psychosocial and physical health factors such as perceptions of child health vulnerability, perceptions of caregiver competence (caregiving satisfaction and self-efficacy/confidence), benefit finding, sleep behaviors, counseling experiences, and symptoms of anxiety and depression that may be related caregiver outcomes. Due to the lack of information available to date, this study aimed to explore these specific aspects of daily living faced by primary caregivers of children diagnosed with SMS in order to examine parental perspectives, behaviors, and current levels of functioning. Four primary research questions were investigated: 1) What rates of anxiety and depression symptoms exist among SMS caregivers and what rates of diminished sleep are reported?, 2) How do perceptions of child health vulnerability, perceptions of caregiver competence (caregiving satisfaction and self-efficacy/confidence), benefit finding, sleep behaviors, counseling experiences, and symptoms of anxiety and depression relate to caregiver well-being?, 3) Do counseling experiences moderate the relationships between anxiety and depression symptoms and caregiver well-being?, and 4) How might these results be used to empower families and educate healthcare providers to address mental health/psychosocial concerns and promote caregiver well-being? It was thought that the results of this study would have implications for ways in which caregivers may benefit from additional psychosocial resources and family-centered care in an effort to promote caregiver well-being more effectively.

## Methods

### Procedure, Participants, and Recruitment to Study

This study was approved by the VCU Institutional Review Board. An exploratory cross-sectional design was utilized, with participants completing a self-report online questionnaire created by a survey development program called *Inquisite*. In addition to the reliable, well-validated measures included in the online questionnaire (see Instrumentation section for details), participants were asked to provide responses to the following open-ended question surrounding the caregiving role: Do you have any other thoughts about your experiences caring for a child diagnosed with Smith-Magenis syndrome that you would like to share at this time? Common themes and quotes from these responses are presented in addition to the quantitative data.

Potential participants included primary caregivers of individuals diagnosed with SMS; the caregivers were at

least 18 years of age and English speaking due to the nature of the questionnaires. Introductory e-mails were sent through the PRISMS (Parents and Researchers Interested in Smith-Magenis Syndrome) listserv and Yahoo SMS Listserv. This email informed potential participants of project objectives, consent procedures, and offered a web link to an online survey if they chose to participate and the option to request a paper version of the questionnaire as an alternate means of participation. Recruitment also took place at the PRISMS support group conference in May 2007 (Reston, VA). All data were collected from May–September 2007. Regardless of how or where participation was completed, participants were fully informed of the research questions under investigation including specific factors (e.g., anxiety, depression symptoms) being analyzed. All data were collected anonymously, meaning that participants did not provide their names or the names of their children at any point during the online questionnaire.

Participants included 115 primary caregivers (i.e., mothers, fathers, aunts, grandmothers) of individuals diagnosed with SMS. Of these, 112 were self-identified as mothers or fathers and are included in the analyses presented within this article. The remaining 3 participants, who reported being aunts or grandmothers, were not included due to the small sample size. Because the questionnaires were completed anonymously online, there is no way to know whether the 112 mothers and fathers are caregivers for 112 different individuals with SMS. It is possible that some of the mothers and fathers were referring to the same child; however, given that the experiences of two parents caring for a single child may be quite different, it seems appropriate to utilize data from two parents who may be referring to the same child. Initial analyses revealed significant differences in caregiver well-being with mothers reporting higher levels of well-being than fathers; therefore, analyses were conducted separately for mothers and fathers (see Table 1 for demographic information comparison). Due to the small number of fathers participating in the study and the inherent problems related to limited power when assessing paternal results, only demographic and correlational data are presented.

#### Caregiver Demographics

Gender, race/ethnicity, age, level of education achieved, income, marital status, number of children in family, hours of sleep obtained per night, hours of sleep desired per night, age of child with SMS, utilization of mental health services, and caregiver physical/mental/dental health care were assessed.

#### Maternal Demographics

A total of 97 mothers participated ( $M_{\text{age}}=41.36$  years,  $SD_{\text{age}}=9.60$  years); see Table 1. Of participating mothers,

**Table 1** Participant Demographics

Variable	Mothers n (%)	Fathers n (%)
<b>Race/ethnicity</b>		
Caucasian/White	91 (94)	15 (100)
Latino/Hispanic	3 (3)	0 (0)
African American/Black	2 (2)	0 (0)
Biracial/Multiracial	1 (1)	0 (0)
<b>Highest level of education completed<sup>a</sup></b>		
High school	7 (7)	2 (15)
Some college	29 (30)	5 (39)
Associate's degree	14 (15)	1 (8)
Bachelor's degree	25 (26)	2 (15)
Some graduate school	6 (6)	0 (0)
Graduate/professional degree	15 (16)	3 (23)
<b>Annual household income<sup>b</sup></b>		
Less than \$15,000	2 (2)	0 (0)
\$15,000 – \$24,999	3 (3)	1 (7)
\$25,000 – \$34,999	6 (6)	0 (0)
\$35,000 – \$44,999	11 (12)	0 (0)
\$45,000 – \$59,999	13 (14)	3 (20)
More than \$60,000	59 (63)	11 (73)
<b>Marital status</b>		
Married	75 (77)	15 (100)
Single	5 (5)	0 (0)
Divorced	12 (13)	0 (0)
Separated	1 (1)	0 (0)
Living with parent of child w/SMS	3 (3)	0 (0)
In contact with other parent	1 (1)	0 (0)
<b>Annual medical exams<sup>a</sup></b>		
Yes	66 (68)	10 (67)
No	30 (31)	5 (33)
<b>Annual dental exams</b>		
Yes	70 (72)	9 (60)
No	27 (28)	6 (40)
<b>Annual eye exams<sup>a</sup></b>		
Yes	40 (41)	6 (40)
No	56 (58)	9 (60)
<b>Anxiety/depression counseling in the past year</b>		
Yes	31 (32)	3 (20)
No	66 (68)	12 (80)
<b>Received anxiety/depression counseling prior to beginning the caregiver role</b>		
Yes	18 (19)	1 (7)
No	79 (81)	14 (93)
<b>Received anxiety/depression counseling after beginning the caregiver role</b>		
Yes	58 (60)	6 (40)
No	39 (40)	9 (60)
<b>Perceives benefits of the caregiving role</b>		
Yes	93 (96)	13 (87)
No	4 (4)	2 (13)

**Table 1** (Continued)

Variable	Mothers n (%)	Fathers n (%)
Specific benefits		
Closer family relationships	51 (53)	9 (60)
Appreciating the preciousness of the child	83 (87)	12 (80)
Having a better perspective on life	81 (84)	12 (80)
Spiritual growth	63 (65)	4 (27)
Learning something about yourself (personal/emotional growth)	74 (76)	10 (67)
Receives support from spouse/partner	78 (80)	14 (93)
Receives support from family members	75 (77)	9 (60)
Receives support from friends	53 (55)	6 (40)
Receives support from		
Teachers/school system	57 (59)	7 (47)
Receives support from Medical professionals	59 (61)	6 (40)
Receives support from		
Mental health professionals	25 (26)	4 (27)
Receives support from social workers	27 (28)	4 (27)
Has other children <sup>a</sup>	82 (86)	12 (80)
Number of other children <sup>a</sup>		
Zero	13 (13)	3 (20)
One	40 (41)	6 (40)
Two	26 (27)	4 (27)
Three	10 (10)	2 (13)
More than three	7 (7)	0 (0)

$n_{\text{mothers}}=97$

<sup>a</sup> 1 mother chose not to answer this question

<sup>b</sup> 3 mothers chose not to answer this question

$n_{\text{fathers}}=15$

93.8% were White, 92.7% had attended at least some college, 62.8% had a household income of more than \$60,000 per year, and 77.3% were married. Only 18.5% had received counseling for anxiety or depressive symptoms prior to the birth of the child with SMS, while 59.8% had engaged in counseling after beginning the caregiver role. On a scale from 1 to 10, with 10 being the most stressful experience of one's life, mothers rated having a child with SMS as 8.02. With respect to how much control or responsibility they feel for their child's genetic syndrome, mothers reported an average score of 6.41 out of 10. Despite challenges, 95.9% of mothers reported perceiving benefits related to experiences caring for a child with SMS.

#### Paternal Demographics

A total of 15 fathers participated ( $M_{\text{age}}=42.07$  years,  $SD_{\text{age}}=9.85$  years); see Table 1. Of fathers participating, 100% were White, 84.6% had attended at least some college, 73.3% had

a household income of more than \$60,000 per year, and 100% were married. Overall, only 6.7% had received counseling for anxiety or depressive symptoms prior to the birth of the child with SMS, while 40.0% had engaged in counseling after beginning the caregiver role. On a scale from 1 to 10, with 10 being the most stressful experience of one's life, fathers rated having a child with SMS as an 8.86. With respect to how much control or responsibility they feel for their child's genetic syndrome, fathers reported an average score of 6.71 out of 10; however, 86.7% of fathers reported perceiving benefits related to his experience caring for a child with SMS.

#### Child Demographics

Caregivers provided demographic information on their children diagnosed with SMS (Table 2). Among their children diagnosed with SMS, mothers reported an average age of 12.91 years ( $SD=9.04$ ), while fathers reported a mean of 11.73 years ( $SD=7.08$ ). Participants reported their children diagnosed with SMS were predominantly female (58% of mothers' reports, 53% of fathers' reports) and Caucasian/White (90% of mothers', 87% of fathers' reports). Nearly all of the paternal participants reported that their child received special school accommodations (93%), while only 75% of maternal participants reported their child received such accommodations. Two thirds of fathers and 82% of mother reported that their child was functioning similarly to or better than other children diagnosed with SMS. See Table 2 for additional demographic comparisons.

#### Instrumentation

##### Vulnerable Child Scale (VCS)

The VCS (Perrin et al. 1989) consists of 14 4-point items ( $1 =$  definitely true,  $4 =$  definitely false) that are summed for a total score with higher scores equating to lower perceived health vulnerability. Statements express various concerns about a child's health or well-being. The VCS has been utilized to assess perceptions of health vulnerability among various populations including parents of infants born prematurely, parents of children diagnosed with cancer, and depressed mothers of preschool-aged children (Anthony et al., 2003; Bendell et al., 1994; Forsyth et al., 1996; Stern et al. 2006). Sample items included: "I often have to keep my child indoors because of health reasons" and "I often think about calling my doctor about my child." The VCS has been reported to have adequate internal consistency reliability (Cronbach's  $\alpha=.75$ ) and excellent test-retest reliability ( $r=.95$ ) (Perrin et al. 1989). Cronbach's  $\alpha$  for this study was .80.

**Table 2** Individual Diagnosed with SMS Demographic Variables

Variable	Mothers' reports n (%)	Fathers' reports n (%)
Gender <sup>a</sup>		
Female	56 (58)	8 (53)
Male	40 (41)	7 (47)
Race/ethnicity <sup>a</sup>		
Caucasian/White	86 (90)	13 (87)
Latino/Hispanic	3 (3)	0 (0)
African American/Black	2 (2)	0 (0)
Asian American	1 (1)	0 (0)
Biracial/Multiracial	4 (4)	2 (13)
Receives special school accommodations		
Yes	73 (75)	14 (93)
No	24 (25)	1 (7)
Comparison to other children with SMS <sup>b</sup>		
Worse than average	17 (18)	5 (33)
About average	33 (36)	5 (33)
Better than average	43 (46)	5 (33)

*n*<sub>mothers</sub>=97<sup>a</sup> 1 mother chose not to answer this question<sup>b</sup> 4 mothers chose not to answer this question*n*<sub>fathers</sub>=15

### Parental Sense of Competence Scale (PSOC)

The PSOC (Gibaud-Wattston and Wandersman 1978) contains 17 items answered on a 6-point Likert scale (1 = strongly disagree, 6 = strongly agree) that assess parenting satisfaction and self-efficacy. Higher scores indicate greater parenting self-esteem. Wording was modified from mother or father to reflect a more general caretaker role. Sample items included: "Being a caretaker is manageable, and any problems are easily solved" and "I honestly believe I have all the skills necessary to be a good caretaker to my child." The PSOC has been utilized to assess parenting competency among parents of typically developing children as well as parents of children with intellectual disabilities (Hassall et al. 2005; Johnston and Mash, 1989). High internal consistencies have been reported for the subscales ( $\alpha$  coefficient of .82 for satisfaction and .70 for self-efficacy) (Gibaud-Wattston and Wandersman 1978). Cronbach's  $\alpha$  for the current study was .72 for caregiver satisfaction and .76 for caregiver self-efficacy.

### Benefit Finding

A revised version of the benefit finding subscale of Mohr and colleagues' Psychosocial Impact Scale was used to assess perceptions of benefits among caregivers (Mohr et al. 1999). Originally normed on patients with multiple sclerosis (MS) and later successfully cross validated with patients diagnosed with cancer and lupus, this 19-item measure asked participants to indicate the extent to which they agree with statements on a Likert scale of 1 (strongly disagree) to 5 (strongly agree) (Katz et al. 2001). In order to

accommodate parents of a child diagnosed with SMS, item stems related to MS were modified slightly. For example, an item that read, "Since having MS, I have learned to communicate better with people" was modified as follows: "Since I began my caregiving role, I have learned to communicate better with people." Additional sample items included: "Caring for my child with SMS has helped me be closer to my family" and "Caring for a child with SMS made me appreciate my life more." Internal consistencies were high, ranging from .79 to .89. Cronbach's  $\alpha$  for this study was .85.

### Center for Epidemiologic Studies—Depression scale (CES-D)

The CES-D is a 20-item global scale developed to measure depressive symptomology in the general population (Radloff 1977). A Likert scale for each item is scored from 0 (rarely or none of the time) to 3 (most or all of the time) to produce a range of possible scores from 0 to 60. Scores ranging from 0–15 indicate "minimum" depressive symptoms, 16–23 "mild" depressive symptoms, and  $\geq 24$  "moderate/severe" depressive symptoms (Rushton et al. 2002). Sample items included: "I felt that I was just as good as other people" and "I felt hopeful about the future." Reliability and validity of the scale has been tested in both general and clinical populations, yielding very good internal consistency for the general population ( $\alpha=.85$ ) and a psychiatric population ( $\alpha=.90$ ) (Radloff 1977). Acceptable discriminate and concurrent validity have been reported for the CES-D using community and psychiatric samples (Weissman et al 1977). Cronbach's  $\alpha$  for the current study was .93.

### *Beck Anxiety Inventory (BAI)*

The Beck Anxiety Inventory (BAI) (Beck et al. 1993) was used to assess anxiety symptomology, and consists of 21 items describing common symptoms of anxiety such as “unable to relax” and “fear of losing control,” with participants indicating the extent to which each item has bothered them over the past week on a Likert scale ranging from 0 (not at all) to 3 (severely, it bothered me a lot). Total scores range from 0 to 63. Scores of 0 to 21 indicate low anxiety, 22 to 35 moderate anxiety, and 36 or more high anxiety. The scale has good internal consistency ( $\alpha=.92$ ) and a one-week retest reliability of .75, with Cronbach’s  $\alpha$  at .89 for this study. Adequate validity has been well-documented (Contreras et al., 2004).

### *Caregiver Well-Being Scale*

Tebb’s (1995) Caregiver Well-Being Scale (Tebb 1995) is built on a health-strengths model (Weick 1986) that asks participants to rate their level of well-being on a Likert scale of 1 (never) to 5 (always). The scale was created based upon a sample of individuals caring for adults or children who were either disabled, severely developmentally delayed, or chronically ill and caregivers of typically developing children. Sample items included: “I feel that I am able to express laughter and joy” and “I feel that I am able to attend to medical and dental needs.” Berg-Weger et al. (2000) reported an internal consistency of .91 for the basic needs subscale (22 items) and an internal consistency of .81 for the activities of living subscale (23 items) (Berg-Weger et al. 2000). Overall reliability was .94. Convergent and discriminant validity were verified with a correlation of .69 between the two subscales. Construct validity is established through factor analysis while criterion validity is supported through the concurrent validity of this measure to a measure of depression. Cronbach’s  $\alpha$  for the current study was .95.

### Data Analyses

This study aimed to explore these specific aspects of daily living faced by primary caregivers of children diagnosed with SMS in order to examine parental perspectives, behaviors, and current levels of functioning. Correlations, hierarchical regression analyses, and identification of common themes associated with qualitative responses to an open-ended question on caregiving roles were utilized to explore ways in which child health vulnerability, perceptions of caregiver competence (caregiving satisfaction and self-efficacy/confidence), benefit finding, sleep behaviors, counseling experiences, and symptoms of anxiety and depression may relate to caregiver well-being. Baron and Kenny’s (1986) interaction

model using a series of hierarchical regression analyses was further utilized to determine whether counseling experiences moderate the relationships between anxiety and depression symptoms and caregiver well-being.

## Results

### Parental Comparison of Sleep Behaviors and Depression and Anxiety Symptoms

Participants in this study reported numerous symptoms related to anxiety and depression; difficulties with sleep behaviors were common, as well. Overall, the majority of caregivers (87.8% of mothers and 92.3% of fathers) reported moderate to severe symptoms of depression. Data also showed that 93.3% of mothers and 100% of fathers assessed reported moderate to high levels of anxiety. Maternal caregivers reported that their sleep was interrupted 4.99 nights/week and that they receive an average total of 6.40 hours of sleep per night. Mothers reported they needed 6.90 hours of sleep/night to function well and ideally they would receive 7.39 hours of sleep/night. Paternal caregivers reported that their sleep was interrupted 5.33 nights/week and that they receive an average total of 4.83 hours of sleep/night. Fathers reported they needed 6.31 hours of sleep per night to function well and ideally they would receive 6.54 hours of sleep/night.

### Relationships Among Demographic and Psychosocial Variables and Caregiver Well-Being

Table 3 provides sample sizes, means, standard deviations, and ranges for all of the primary psychosocial variables under investigation, including perceived child vulnerability, caregiver satisfaction, caregiver self-efficacy, benefit finding, depression, anxiety, and caregiver well-being.

### *Correlational Data Related to Caregiver Well-Being*

*Maternal Participants* Among mothers participating in the study, those with higher levels of formal education ( $r=.29$ ,  $p<.01$ ) indicated greater caregiver well-being. Mothers who reported having received counseling for depression or anxiety after beginning their caregiving role were more likely to show higher levels of caregiver well-being ( $r=-.45$ ,  $p<.001$ ). Higher levels of caregiver well-being were also related to lower levels of perceived child health vulnerability ( $r=.36$ ,  $p<.001$ ), greater satisfaction with the caregiving role ( $r=.60$ ,  $p<.001$ ), higher caregiving self-efficacy ( $r=.43$ ,  $p<.001$ ), increased benefit finding ( $r=.22$ ,  $p<.05$ ), and lower endorsement of symptoms of anxiety

**Table 3** Psychosocial Variable Means, Standard Deviations, Sample Sizes, and Ranges

Variable	Mothers				Fathers			
	M	SD	n (%)	Range	M	SD	n (%)	Range
Perceived child vulnerability	38.8	7.2	96 (99%)	24.0–54.0	39.0	5.8	15 (100%)	27.0–49.0
Parenting satisfaction	36.0	6.8	95 (98%)	22.0–51.0	33.2	5.4	15 (100%)	23.0–42.0
Parenting self-efficacy	28.4	5.5	96 (99%)	15.0–41.0	27.6	4.9	15 (100%)	19.0–33.0
Benefit finding	66.8	9.6	94 (97%)	33.0–84.0	58.1	13.8	14 (93%)	30.0–84.0
Depression	36.7	11.6	90 (93%)	20.0–72.0	43.7	12.2	13 (87%)	25.0–65.0
Anxiety	30.9	8.5	90 (93%)	21.0–55.0	31.8	9.2	13 (87%)	21.0–56.0
Caregiver well-being	154.0	27.0	90 (93%)	81.0–225.0	135.8	21.5	13 (87%)	102.0–187.0

*M* mean, *SD* standard deviation, *n* number of participants

Highest possible *n* for mothers was 97; Highest possible *n* for fathers was 15

( $r=-.54$ ,  $p<.001$ ) and depression ( $r=-.76$ ,  $p<.001$ ). See Table 4 for additional information.

**Paternal Participants** Among fathers participating in the study, those who reported more benefits of having a child diagnosed with SMS were more likely to endorse higher levels of caregiver well-being ( $r=.67$ ,  $p<.01$ ). Those who indicated fewer symptoms of depression also reported higher levels of caregiver well-being ( $r=-.63$ ,  $p<.02$ ). See Table 5 for additional information.

#### *Effects of Perceived Child Vulnerability, Caregiver Satisfaction and Self-Efficacy, and Benefit Finding on Maternal Caregiver Well-Being*

Hierarchical regression analyses were utilized to assess the direct effects of perceived child vulnerability, caregiver satisfaction, caregiver self-efficacy, and benefit finding on maternal caregiver well-being (Table 6). As stated, due to a small sample size ( $n=15$ ), no regression analyses were conducted utilizing the fathers' data. Among mothers, based on significant correlations, highest level of education achieved was added as a covariate. The entire model was found to be significant ( $F(5, 82)=16.97$ ,  $p<.001$ ) and

accounted for 51% of the variance in caregiver well-being ( $R^2=.51$ ). Perceived child vulnerability, caregiver satisfaction, caregiver self-efficacy, and benefit finding were significantly related to the changes in caregiver well-being beyond the influence of education ( $\Delta F(4, 82)=17.61$ ,  $p<.001$ ). Together, perceived child vulnerability, caregiver satisfaction, caregiver self-efficacy, and benefit finding accounted for 42% of the variance in caregiver well-being ( $\Delta R^2=.42$ ). Independently, caregiver well-being was directly influenced by perceived child vulnerability ( $B=.75$ ,  $p<.02$ ), which accounted for 3.8% of the unique variance in caregiver well-being ( $sr^2=.038$ ), caregiver satisfaction ( $B=1.78$ ,  $p<.001$ ), which accounted for 16.4% of the unique variance in caregiver well-being ( $sr^2=.164$ ), and benefit finding ( $B=.49$ ,  $p<.04$ ), which accounted for 2.6% of the unique variance in caregiver well-being ( $sr^2=.026$ ).

#### *Interaction Between Counseling Experiences and Anxiety or Depression Symptoms on Caregiver Well-Being*

Baron and Kenny's (1986) regression model for exploring moderations was utilized to determine interaction effects between anxiety or depressive symptomatology and anxiety/depression counseling after beginning the caregiver role

**Table 4** Intercorrelations among Maternal Psychosocial Variables

Variable	1	2	3	4	5	6	7
1 Perceived vulnerability	–						
2 Caregiver satisfaction	.28**	–					
3 Caregiver efficacy	.28**	.39***	–				
4 Benefit finding	-.06	.12	.19	–			
5 Anxiety	-.33***	-.30**	-.25*	-.17	–		
6 Depression	-.45***	-.58***	-.46***	-.23*	.68***	–	
7 Caregiver well-being	.36***	.60***	.43***	.21*	-.54***	-.76***	–

$n=97$ ; \*  $p<.05$ , \*\*  $p<.01$ , \*\*\*  $p<.001$

**Table 5** Intercorrelations among Paternal Psychosocial Variables

Variable	1	2	3	4	5	6	7
1 Perceived vulnerability	–						
2 Caregiver satisfaction	.33	–					
3 Caregiver efficacy	.10	.62*	–				
4 Benefit finding	-.36	-.38	-.07	–			
5 Anxiety	.15	-.42	-.42	-.15	–		
6 Depression	.07	-.14	-.34	-.27	.35	–	
7 Caregiver well-being	.22	-.11	.12	.64*	-.15	-.56*	–

$n=15$ ; \*  $p<.05$ , \*\*  $p<.01$ , \*\*\*  $p<.001$

on overall caregiver well-being (Baron and Kenny 1986). A significant moderating effect of depression/anxiety counseling after beginning the caregiver role on the relationship between anxiety symptomatology and caregiver well-being was observed (overall model:  $F(4, 84)=14.79$ ,  $p<.001$ ; interaction term:  $F(1, 84)=7.94$ ,  $B=1.97$ ,  $R^2=.06$ ,  $p<.01$ ) with the entire model accounting for 41% of the variance in caregiver well-being ( $R^2=.41$ ) and the interaction uniquely accounting for 5.5% of the variance ( $sr^2=.055$ ). Results suggest that those who report lower levels of anxiety and do not seek counseling report the highest levels of caregiver well-being (Table 7, Fig. 1). Those who show high levels of anxiety but do not seek counseling fair the worst in terms of well-being. There was no moderating effect of depression/anxiety counseling after beginning the caregiver role on the relationship between depressive symptomatology and caregiver well-being (Table 8).

#### Parents' Descriptions of Caring for a Child Diagnosed with SMS

Participants were given the opportunity to provide additional thoughts on what it is like to care for a child diagnosed with SMS. Of the 112 parent participants, 73 (65%) chose to write about their experiences in this open-ended format. These comments provided honest insight regarding the daily challenges, benefits, accomplishments, ongoing concerns, and personal struggles of this caregiving role. Responses were reviewed, and common themes were

identified. Among the 73 responses, the most common themes discussed included the following: (1) Benefit finding/acceptance/gratitude/personal growth (41.1%; i.e., seeing positive aspects of having a child with SMS); (2) Behavioral issues (32.9%; i.e., aggression, self-injurious behaviors, tantrums); (3) Coping and adaptation (30.1%; i.e., coping with daily challenges related to having a child with SMS and/or discussion of being better able to cope as the child aged); (4) Emotional distress or anxiety (20.5%); (5) Social support (19.2%; i.e., struggling to receive adequate support); (6) The need for public awareness, policy, and literature (17.8%); (7) Family dynamics/marriage (13.9%; i.e., difficulties with meeting the entire family's needs and maintaining family relationships); (8) Sibling issues (13.7%; i.e., parents concerns about adaptation and coping among non-SMS siblings); and (9) Parenting skills (13.7%; i.e., concerns that parenting skills are insufficient). Additional themes (reported by less than 10% of parents) included normalcy (i.e., the desire to lead a typical home life and have children with SMS treated as typically developing children); diagnosis (i.e., struggling to diagnose SMS and/or feeling relieved following the diagnosis); sleep issues; parental career/educational concerns; feelings of isolation; sacrifices made due to the caregiving role; counseling; relationships with God; attachment issues; school issues for the child with SMS; and concerns about the future and helping the child with SMS develop independent living skills. A representative sample of caregiver quotes is provided in Table 9.

**Table 6** Summary of Hierarchical Regression Analysis for Influences of Perceived Vulnerability, Caregiver Satisfaction, Caregiver Self-efficacy, and Benefit Finding on Caregiver Well-being among Mothers

Variable	B	SE B	$\beta$	$sr^2$
Step 1				
Highest level of education completed	5.28**	1.85	.29**	.08**
Step 2				
Highest level of education completed	4.43**	1.43	.25**	.06**
Perceived vulnerability	.75*	.30	.21*	.04*
Caregiver satisfaction	1.78***	.34	.45***	.16***
Caregiver self-efficacy	.63	.43	.13	.01
Benefit finding	.49*	.23	.17*	.03*

$R^2=.09$  for Step 1 ( $p=.006$ );  $\Delta R^2=.42$  for Step 2 ( $p<.001$ ). \* $p<.05$ , \*\* $p<.01$ , \*\*\* $p<.001$

**Table 7** Summary of Hierarchical Regression Analyses for Moderation of Counseling after Beginning the Caregiving Role on the Relationship between Anxiety Symptomatology and Caregiver Well-being among Mothers

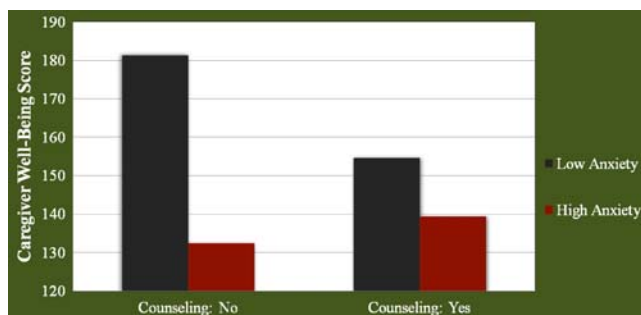
Variable	B	SE B	$\beta$	$sr^2$
Step 1				
Highest level of education completed	5.22**	1.86	.29**	.08**
Step 2				
Highest level of education completed	1.79	1.77	.10	.01
Anxiety	-1.59***	.31	-.50***	.21***
Step 3				
Highest level of education obtained	1.16	1.71	.05	.00
Anxiety	-1.30***	.31	-.41***	.13***
Counseling	-14.60**	5.18	-.27**	.06**
Step 4				
Highest level of education obtained	.92	1.65	.05	.00
Anxiety	-4.83***	1.29	-1.52***	.10***
Counseling	-9.82	5.26	-.18	.03
Anxiety X counseling	1.97**	.70	1.10**	.05***

$R^2 = .08$  for Step 1 ( $p = .006$ );  $\Delta R^2 = .21$  for Step 2 ( $p < .001$ );  $\Delta R^2 = .06$  for Step 3 ( $p = .006$ );  $\Delta R^2 = .06$  for Step 4 ( $p = .006$ ). \* $p < .05$ , \*\* $p < .01$ , \*\*\* $p < .001$

## Discussion

### Major Research Findings

The results of this study suggest that primary caregivers of children with Smith-Magenis syndrome experience increased amounts of distress within the caregiving role in terms of depression and anxiety symptoms, sleep behaviors, and their relations to caregiver well-being. Although 40% of fathers and approximately 60% of mothers sought out counseling services after beginning the caregiving role, symptoms of anxiety and depression continued to persist. A number of factors appear to play roles in determining how well SMS caregivers are coping with and adjusting to daily caregiving demands. Among mothers, important factors related to improved caregiving include being satisfied with the caregiving role, having confidence in one's caregiving abilities (i.e., caregiver self-efficacy), having a realistic view of her child's health vulnerabilities, and perceiving



**Fig. 1** Counseling Improves Caregiver Well-being among Highly Anxious Caregivers. Interaction Graph Showing Relationships between Counseling History Following Initiation of the Caregiving Role and Current Anxiety Symptoms on Caregiver Well-being. Caregivers Who Show High Levels of Anxiety but Do Not Seek Counseling Fair the Worst in Terms of Well-being.

benefits related to the caregiving role. For those with the highest levels of anxiety symptoms, counseling seems to help to alleviate these symptoms and to improve caregiver well-being. Interestingly, results of the interaction analysis specifically show that those mothers with fewer symptoms of anxiety may not benefit as much in terms of caregiver well-being when counseling is sought. These findings suggest the need for additional screening procedures that would better identify which parents may be in need of specific counseling services, discussing the many options for therapeutic intervention with the caregivers, and then offering the most appropriate services and resources based on the level of distress and the needs of the family. It is possible that mothers reporting fewer symptoms of anxiety may benefit more from different types of interventions that were not specifically assessed in this study such as support groups, home-based counseling services, and/or respite care.

Due to the small number of fathers who participated in this study, definitive results are difficult to address. Trends in the data seem to indicate that fathers who are able to perceive more gains and benefits of having a child with special needs also have higher levels of well-being related to the caregiving role. Reduction of depressive symptoms also seems to be related to greater well-being.

Overall, parents appear to vary in their perceptions of how they are coping with and embracing their caregiving roles. Quotes from parents echo the results of the quantitative analyses, with most parents reporting many benefits and advantages of coping with a child diagnosed with SMS despite the ongoing obstacles, frustrations, and daily worries. Being able to perceive benefits of having a child with SMS seems to be especially important among both mothers and fathers in terms of promoting caregiver well-being.

Although there is limited research concerning caregiver well-being among parents caring for a child with SMS,

**Table 8** Summary of Hierarchical Regression Analyses for Moderation of Counseling after Beginning the Caregiving Role on the Relationship between Depression Symptomatology and Caregiver Well-being among Mothers

Variable	B	SE B	$\beta$	$sr^2$
Step 1				
Highest level of education completed	5.22**	1.86	.29**	.08**
Step 2				
Highest level of education completed	1.48	1.32	.08	.01
Depression	-1.70***	.17	-.73***	.49***
Step 3				
Highest level of education obtained	1.23	1.33	.07	.00
Depression	-1.60***	.19	-.69***	.36***
Counseling	-5.44	4.35	-.10	.01
Step 4				
Highest level of education obtained	1.23	1.32	.07	.00
Depression	-2.67***	.66	-1.15***	.08***
Counseling	-3.70	4.43	-.07	.00
Depression X counseling	.65	.38	.46	.01

$R^2 = .08$  for Step 1 ( $p = .006$ );  $\Delta R^2 = .50$  for Step 2 ( $p < .001$ );  $\Delta R^2 = .01$  for Step 3 ( $p = ns$ );  $\Delta R^2 = .01$  for Step 4 ( $p = ns$ ).  
\* $p < .05$ , \*\* $p < .01$ , \*\*\* $p < .001$

**Table 9** Representative Parent Comments on Caring for a Child Diagnosed with SMS

- "I have often said I think I needed him much more than he has ever needed me. I have seen such tremendous growth in my other children. My son is amazing except when he's not. I tend to separate him from the disorder meaning [my child] is not SMS; he is a loving, productive, essential part of our family who happens to have this disorder called SMS. He had an unfortunate luck of the draw, but I believe we all have lessons to learn and in some way this disorder is our family's way of learning our lessons. I am a profoundly different person for having [him] in my life. It's certainly not to say that it hasn't been incredibly difficult at times; it has. But I believe there is no sense on losing yourself [for something] essentially you have no control over. We do the best we can each day. Some days we get through with a little more grace than others."
- "It is a lonely, isolating proposition because most people cannot grasp how extremely difficult and tiring raising an SMS child is."
- "Terror of what would happen to her in the future motivated me to continue even when it seemed hopeless...which was often."
- "I love my daughter very much and she means the world to me. I was just amazed at the fact that many of the parents I met...seemed to have a different look on the whole situation than I do...But I personally just don't see how my daughter having a disorder is a blessing...I have also had a pretty stressful life and I always hear "it's a test of faith." Well how many tests can I possibly have? I know none of these children deserve this and don't know why it had to happen."
- "Not knowing your child has SMS is by far worse than knowing. You go thru so many years of finger pointing and terrible accusations by education professionals and receive a lot of criticism regarding your parenting skills. Your self-esteem really takes a beating and every answer you have for people gets reflected back at you as only an excuse or a cover up of some sort. It is very hurtful and demeaning to hear these things. I did every therapy available to my child and sacrificed a lot. When I found out the real diagnosis and became an overnight professional on SMS. My inner self was absolutely set free, and the Mom who knew her child was finally out and ready to rub some noses into it. I can now argue on his behalf and feel confident that I know who he is and what he needs and will fight till the end to get what he needs...It's very much been a life changing and positive in our lives as odd as that may sound."
- "This condition awakens the mind to GO BEYOND lateral thinking when anticipating SMS behavioural characteristics. Learning about SMS has been difficult, with little or no information readily available, and in my region there are not real individual organizations or doctors with a vast knowledge of SMS. Although sites such as PRISMS are a godsend, my main difficulty or frustration is feeling helpless on occasions not being able to stop the self harm behaviour. My daughter means the world too me, and I worry about the future, what it holds for her. SMS children are most loving and wonderful. As a father of a beautiful little girl with SMS, I have experienced the highs and lows of this rare disorder."
- "Although there are moments when it is difficult to care for my son, I draw from his strength. He has been through so much in his short life, yet he always has a smile for me."
- "SMS is an ongoing "onion" that has so many complex layers that continually unravel. When one behavior settles down, another one emerges, and you never know what is coming next. There are still so many unknowns with this disorder, and many uncertainties apply."
- "It is by far the hardest thing I do...all day, every day...it's life consuming and life draining. It's also rewarding. Every little accomplishment, from our SMS daughter but also our non-SMS daughter is treasured. We value the things that need to be valued."
- "This is the toughest job I can ever imagine. Sometimes I resent other 'typically developing families' with their insignificant challenges. However, I always remember that it could be worse and remind myself that GOD trusted me with this child. My motto is 'If you're handed it...you can handle it.'"

Seventy-three parents provided written comments

results are comparable to the available extant literature on SMS and genetic syndromes with similar physical, emotional, and behavioral symptoms. According to Hodapp et al. (1998) and Fidler et al. (2000), the best predictor of pessimism among parents was the level of maladaptive behaviors displayed by the children diagnosed with SMS. Greater stress was reported among parents of children with greater levels of developmental disabilities (Hodapp et al. 1998), and parents of children with SMS reported greater pessimism and parent and family problems than those families coping with Down syndrome (Fidler et al. 2000). Social support appeared to be an important moderating factor in stress level outcomes. The current study found counseling experiences, which is considered a form of support, to be an important moderating factor related to caregiver well-being. Benefit finding, a construct somewhat opposite of pessimism, which has been reported to be an important factor in helping individuals cope with chronic illness (Katz et al. 2001), was identified as being very salient in its relation to caregiver well-being with greater levels of benefit finding being related to greater caregiver well-being. Results of this study were also similar to those of van den Borne and colleagues (2006) who reported relatively high levels of depression symptoms and fears about the future for themselves and their children among parents caring for children with Prader-Willi or Angelman syndrome. Additional studies on caregiver well-being among those with SMS and other genetic syndromes will offer additional insight into stressors affecting these families negatively and positive factors such as benefit-finding that may serve as a means of promoting coping and adjustment.

### Practical Implications

While it is critical for caregivers to be aware of their increased risk to experience symptoms of depression and anxiety, enhanced awareness on the part of genetic counselors and other healthcare providers is also imperative. Each day, genetic counselors present difficult and life-changing information to caregivers and their families. Through this process, it may be easy to lose sight of the needs of the caregivers given the abundant needs of the child and the amount of information that must be provided to the family. Studies such as these may assist genetic counselors in identifying questions to ask caregivers regarding support systems and psychosocial functioning. Given that genetic counselors may be among the first people speaking with the family about the SMS (or other genetic) diagnosis, such professionals are in a pivotal position for offering initial screening to primary caregivers for depression and anxiety symptoms, suggesting referrals for mental health services, and providing information

regarding ongoing support groups and respite services. When available, genetic counselors may be able to network with psychologists and social workers during parent consultations to provide immediate multidisciplinary care to families.

As health care systems continue to move toward family-centered care, all healthcare providers are encouraged to make the mental well-being of caregivers a priority and to make appropriate referrals when necessary. Referrals to mental health specialists may enable caregivers to receive necessary treatment and utilize better coping mechanisms to deal with their depression and anxiety symptoms, improve caregiver self-efficacy and competence, identify benefits of caring for a child with SMS, and accurately identify their child's health vulnerabilities. Aid provided by respite services may enable caregivers to take the time to take better care of themselves. Referrals to support groups may also help caregivers as speaking with individuals in similar circumstances may introduce new coping methods and different support services to caregivers.

### Study Limitations

One limitation of this study was bias within the sample. The sample represented an estimated 20% of the registered parents of PRISMS, the SMS support group. The sample is well-educated, with almost 50% reporting that they have attained a bachelors degree or higher. Furthermore, the majority of participants were married and financially stable. Such individuals are likely to have better access to support services for themselves and their child, yet they continue to suffer from moderate to severe symptoms of depression and anxiety and feel that caring for their child with SMS is one of the most stressful experiences in their lives. Another limitation concerns the small sample of fathers, which, as mentioned previously, precludes drawing definitive conclusions.

### Research Recommendations

Using this study as a model, comparison of these data to the same variables in families of children with similar genetic syndromes, such as Prader-Willi, Angelman, and Williams syndromes, may provide more insight into the complexities of living with a child with a chronic, complex genetic syndrome. Future studies may also benefit from including families who represent broader social and educational strata in order to better determine the needs of individual families and provide recommendations for ongoing psychosocial resources. Understanding the experiences of other family members such as siblings would provide additional insight into the daily functioning and needs of these families.

## Conclusions

Keeping caregivers healthy is essential to enabling them to provide the best care and environment for their children and to support the well-being of the family unit. Seeking counseling may be especially helpful in not only alleviating these symptoms but improving quality of life among caregivers and families. Explaining these increased risks for depression and anxiety to the caregivers at the time of diagnosis may reduce the incidence or severity of mental health issues for these individuals, providing for better quality of life for the entire family. Furthermore, efforts must be made to better prepare healthcare providers to address the mental health needs of primary caregivers and to make appropriate referrals aimed at promoting the well-being of the entire family.

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