




Exploring the impact of pediatric short bowel syndrome on parent well-being using a disease-specific pilot survey

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Abstract

Background: Children with short bowel syndrome (SBS) have complex care needs, most of which are met in the home by family caregivers who may experience a range of stressors unique to this experience. Prior research suggests that parents of children with SBS have poorer health-related quality of life than peers parenting children without health needs, but the mechanisms shaping parent outcomes are understudied.

Methods: A pilot survey was developed using a community-driven research design to measure the impact of disease-specific items on parent-perceived well-being. The cross-sectional survey, which included both closed-ended and open-ended items, was distributed to a convenience sample of parents of children with SBS. Quantitative and qualitative data were integrated for a mixed-methods analysis of how individual items impacted parent well-being.

Results: Twenty parents completed the survey. *Sleep interruptions, lack of support and resources, and psychological stressors* and their mental health implications were more frequently reported as stressors than logistics related to caregiving (e.g., managing therapies and preparing specialized meals).

Conclusion: The impact of a child's SBS on parent well-being may stem mainly from three interconnected domains: poor sleep and its consequences, lack of access to support and resources, and a range of psychological stressors that affect parent mental health. Understanding the mechanisms through which SBS shapes parent well-being is a necessary first step for developing targeted interventions to support parents and provide family-centered care.

KEYWORDS

caregiver burden, enteral nutrition, intestinal failure, parenteral nutrition, pediatrics, quality of life, short bowel syndrome

Marie L. Neumann, Jessica Y. Allen, Amy Ladner, and Swapna Kakani are SBS community stakeholders.

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INTRODUCTION

Children with short bowel syndrome (SBS), the leading cause of intestinal failure (IF), have complex care needs, which often include long-term parenteral nutrition (PN) or enteral nutrition (EN), specialized diets, hormonal therapies, and medical or surgical interventions.^{1,2} Most of these needs are met in the home setting by parents or family caregivers, who may in turn experience stressors unique to their caregiving role. Prior research suggests family caregiving impacts all aspects of family life, including reduced productivity at work, underemployment or unemployment, decreased recreational or social activities, disturbances to familial and social relationships, increased symptoms of depression, ongoing worries about their loved one, and fatigue.^{3–5}

The existing literature points to the complexity of the caregiving experience for parents of children with SBS or IF. The largest published study of parents to children with IF reported health-related quality of life (HRQOL) was lower when compared with parents of healthy children but similar to parents of children with other chronic illnesses.⁶ Interestingly, family functioning scores were *higher* among families of children with IF than among the reference group, suggesting both positive and negative outcomes may be associated with caregiving.⁶ Belza and colleagues found higher levels of stress, anxiety, and depression among parents to children with IF receiving PN.⁷ Interviews with parents revealed they perceived care work as an around-the-clock commitment that requires a high level of skill and vigilance affecting sleep, ability to work and participate in restorative activities, parental roles, and mental and emotional well-being.³ At the same time, parents highlighted their family's resilience and ability to cope with their child's complex needs.³ Similarly, Neumann et al found that families of children with SBS perceived they successfully manage their child's condition with confidence and integrate care into family routines while simultaneously expending a substantial time and effort to do so.⁸

Prior research suggests that parents of children with SBS will experience stressors associated with their child's condition that may result in decreased family HRQOL. Caregiver physical and mental well-being may be related to patient outcomes and, thus, be an important area of intervention.^{9,10} Clinicians, especially those within specialized centers, have called for the incorporation of assessments and supports targeted at caregivers.^{3,4,6,11} However, a lack of disease-specific measures limits the understanding of mechanisms responsible for impaired parent QoL, which would aid in developing targeted interventions. Thus, the objective of this study was to explore the SBS-related factors associated with parent QoL

using a mixed-methods disease-specific pilot survey co-developed by SBS community members and clinicians.

MATERIALS AND METHODS

Study design and participants

The recruitment processes utilized in this study have been previously described elsewhere.⁸ Briefly, the core research team, as well as a project stakeholder committee, consisted of members of the SBS community (patients, caregivers, and advocates). Members of the stakeholder committee who were adult caregivers of minor children (age <18 years) with SBS were eligible to participate in the study. Unique links to a cross-sectional, web-based questionnaire hosted on RED-Cap were emailed to caregivers, with multiple reminder emails sent. The survey was open from March 2021 to May 2021. Respondents provided informed consent prior to beginning the survey. The study was approved by the institution's institutional review board. The final sample consisted of 20 caregivers of children diagnosed with SBS who completed relevant sections of the survey.

Measures

A SBS-specific pilot survey tool was developed to measure both (parent-perceived) child QoL and parent QoL and has been previously described.⁸ The pilot survey included several subsections, including parent and child demographics, child medical background, and child QoL. An additional subsection captured parent perceptions of the negative impact of a matrix of 19 disease-specific items; for each item, parents were asked to indicate the strength of the item's negative impact on their own overall well-being in the past year using a 5-point Likert scale; a "not applicable" option was available for each item. "Well-being" was defined as a "state of being comfortable, healthy, or happy overall." For each item rated with a 4 or a 5 (indicating a strong negative impact), parents were prompted to explain their answers in open-ended follow-up questions. One additional open-ended question asked parents if any other SBS-specific items not included in the matrix had a negative impact on their overall well-being. Lastly, an open-ended question asked parents to describe items that positively impacted their well-being or helped them cope.

Data analysis

Quantitative analyses were conducted using Stata/SE v. 16 (StataCorp LLC). Survey responses were examined for

missing values and normality of distribution; basic descriptive analyses assessed survey and sample characteristics. To determine the proportion of parents who perceived select disease-specific items to have a strong negative impact on their own well-being, the number of respondents who selected a “4” or a “5” on the SBS disease-specific item matrix was divided by the number of respondents who endorsed the item as the denominator. Some survey items were agreed upon by the research team to be applicable to all respondents (eg, toileting, sleep); in these cases, responses indicating an item was “not applicable” were included in the denominator when calculating percentages (Figure 1 note).

Qualitative responses were analyzed for themes related to each disease-specific item utilizing MAXQDA (VERBI Software, 2020) by a primary coder. A secondary coder reviewed these classifications, with interrater reliability at 0.93 between coders. Differences were

resolved through team communication to reach shared consensus. In two cases, the qualitative data were used to clarify or adjust responses to a quantitative item (see Figure 1 note). Quantitative and qualitative data were integrated for a mixed-methods analysis of the ways in which individual items impacted parent well-being.

RESULTS

Parent and child characteristics

Parent and child characteristics are presented in Table 1. The survey sample was comprised largely of highly formally-educated women (80%) and men (20%), and included four adoptive parents and one foster parent. Respondents are referred to collectively as “parents” for consistency and clarity. Of the boys (60%) and girls (40%)

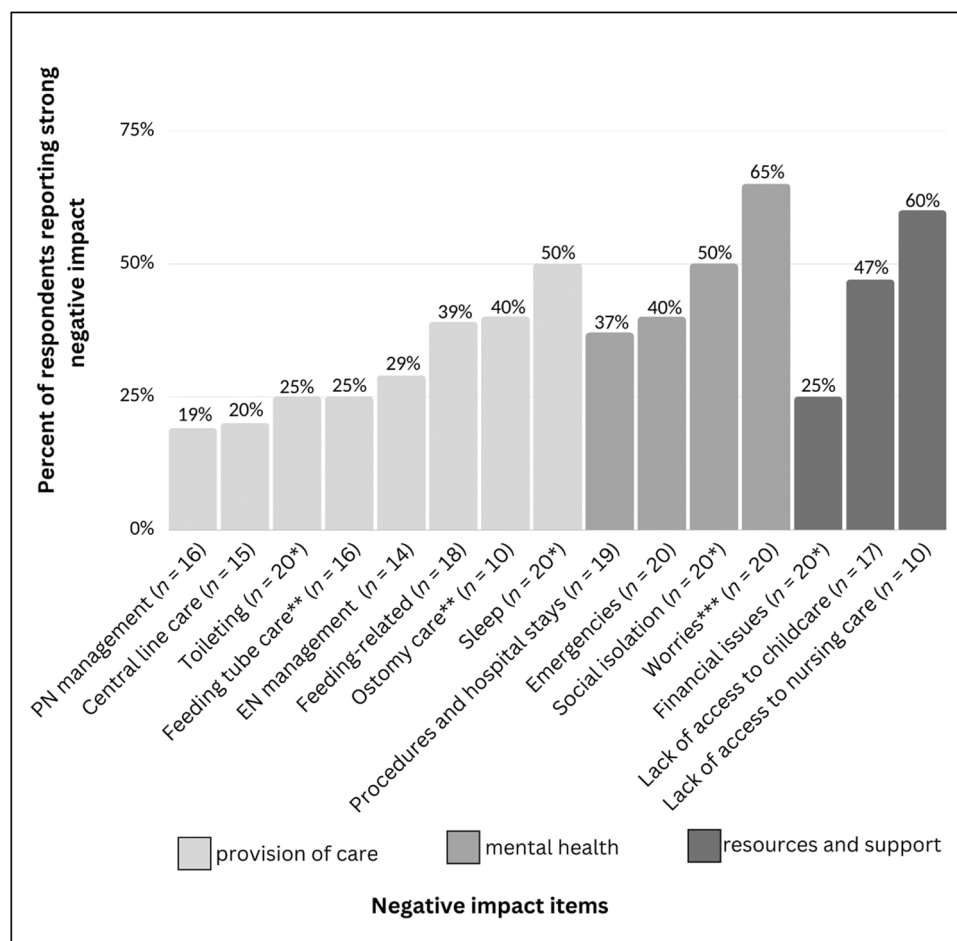


FIGURE 1 A bar graph showing the percent of participants reporting an item as having a strong negative impact on their overall well-being. Percentages were calculated as the number of parents who selected a “4” or a “5” on the short bowel syndrome disease-specific survey tool divided by the number who endorsed each item. *Parent responses indicating an item was “not applicable” were included in the denominator in calculating percentages. **The original combined feeding tube/ostomy care item was retroactively separated into a feeding tube and an ostomy item based on open-ended responses provided by respondents for this item. ***The worries item combines two separate survey items: worries about interactions with peers and worries about the child’s future. EN, enteral nutrition; PN, parenteral nutrition.

TABLE 1 Caregiver and child characteristics (N = 20).

Category	Response	n (%)
Caregiver characteristics		
Relationship to child with SBS	Biological parent	15 (75)
	Adoptive parent	4 (20)
	Foster parent	1 (5)
Sex	Female	16 (80)
	Male	4 (20)
Race/ethnicity	Non-Hispanic White	19 (95)
	Hispanic/Latino	1 (5)
Age, years	19–29	2 (10)
	30–39	5 (25)
	40–49	11 (55)
	50+	2 (10)
Educational background	Some college	3 (15)
	Bachelor's degree	8 (40)
	Advanced degree	9 (45)
Employment status	Working full-time	7 (35)
	Working part-time	5 (25)
	Retired/homemaker/disabled	7 (35)
	Student	1 (5)
Relationship status	Married/cohabiting	18 (90)
	Single	1 (5)
	Divorced	1 (5)
Spiritual identity	Christian	12 (60)
	Jewish	1 (5)
	Not religious	6 (30)
	Prefer not to say	1 (5)
Number of children in household	1	11 (55)
	2–3	9 (45)
SBS child characteristics		
SBS child's sex	Female	8 (40)
	Male	12 (60)
Underlying cause of SBS	NEC	6 (30)
	Gastroschisis	4 (20)
	Hirschsprung's disease	4 (20)
	Jejunal atresia	4 (20)
	Other	2 (10)
	Age at the time of the survey	0–12 months
	Between 1 and 6 years old	7 (35)

TABLE 1 (Continued)

Category	Response	n (%)
	Between 6 and 10 years old	7 (35)
	Between 10 and 14 years old	3 (15)
	Between 14 and 17 years old	1 (5)
Comorbidities secondary to SBS	Yes	11 (55)
	No	9 (45)
PN status	Currently receiving PN	16 (80)
	Previously received PN	4 (20)
	Never received PN	0
EN status	Currently receiving EN	11 (55)
	Previously received EN	7 (35)
	Never received EN	2 (10)
Currently on both PN and EN	Yes	9 (45)
	No	11 (55)
Ostomy status	Currently has ostomy	10 (50)
	Previously had ostomy	5 (25)
	Never had ostomy	4 (20)
	Unsure	1 (5)
SBS-related care managed by IRP	Yes	14 (70)
	No/unsure.	6 (30)

Abbreviations: EN, enteral nutrition; IRP, intestinal rehabilitation program; NEC, necrotizing enterocolitis; PN, parenteral nutrition; SBS, short bowel syndrome.

whose parents participated in the study, a large share was currently receiving PN (80%) or EN (55%), with 45% currently receiving a combination of both; notably, 50% currently had an ostomy. Over half of children (55%) had comorbidities secondary to their SBS, most commonly neurodevelopmental in nature (40%). Most respondents (70%) reported their child's SBS-related care was being managed by an established intestinal rehabilitation program.

Negative impact of disease-specific items on parent overall well-being

Parent responses to select disease-specific survey items are shown in Table 2, with the percentage of respondents reporting a *strong negative impact* of these items on their well-being displayed in Figure 1. Thematic analyses of

TABLE 2 Parent responses to select items in the disease-specific item matrix (n = 20).

Think about your experiences as a caregiver to your child with SBS over the past year. To what extent would you say the following items have had a NEGATIVE impact on YOUR overall well-being (ie your sense of being comfortable, healthy, and happy)?

Disease-specific item	Description	0, not applicable	1, no negative impact	2	3, somewhat of a negative impact	4	5, high negative impact
Your child's PN management	Preparing and hooking up to PN, wearing a backpack, keeping to a PN schedule, etc	4	3	3	7	3	0
Your child's central line care and management ^a	Line infections, placement of catheters, line breaks, site care	4	3	3	6	3	0
Child's enteral feeds	Hooking up on feeds, preparing feeds, etc	6	2	4	4	2	2
Child's feeding tube/ostomy care	Insertion, placement, care, maintenance, leaking and blowouts of ostomy bags, prolapse of stomas	5	0	5	2	6	2
Child toileting	Frequent diaper changes or trips to the toilet, soiled clothing, difficulty potty training, etc	3	5	2	5	2	3
Child's feeding-related issues	Problems with feeding or relationship to food, restrictions on what foods are allowed and limitations on how much	2	3	3	5	4	3
Procedures	Child's medical procedures	1	4	4	7	3	1
Hospitalizations	Child's hospital stays	5	2	3	6	2	2
Sleep interruptions	Sleep interruptions to care for child	2	0	4	4	4	6
Worries: future	Worries about your child's future	1	2	1	3	8	5
Worries: peers	Worries about the child not fitting in with peers	0	3	3	7	3	4
Emergencies	Always having to be prepared for emergencies	0	2	3	7	1	7
Financial issues	Health insurance issues, coverage of procedures or medical supplies, paying for treatment or procedures	1	5	7	2	2	3
Social isolation	Being isolated to keep child from getting sick, having no one else who can care for child	2	2	2	4	6	4
Childcare	Difficulty securing adequate childcare for the child	3	2	3	4	5	3
Nursing	Difficulty securing home nursing care for the child	10	0	1	3	4	2

Abbreviations: PN, parenteral nutrition; SBS, short bowel syndrome.
^an = 19 for this item as one parent failed to provide a response for this item.

the qualitative responses to follow-up questions for each item revealed several subthemes, outlined in Table 3.

Provision of care

Items related to the provision of care for the child with SBS correspond to the management of the child's medical devices or therapies and caring for needs related to feeding or toileting. Of all care provision items, PN management and central line care were least frequently reported (19% and 20%, respectively) as having a strong negative impact on parent well-being, with feeding-related and ostomy-related care most frequently reported (39% and 40%, respectively). Responses to open-ended follow-up questions corresponding to caregiving items revealed two diverging stressors related to care provision: logistical and psychological. *Logistical stressors* such as the preparation of feeds, changing of child's dressing, laundry and cleaning after toileting accidents, and the preparation of meals were highlighted in 10 parent responses:

[My child receives] a mixture of continuous pump feeds and gravity boluses. I'm constantly needing to address something with her feeds or her difficulty tolerating them... stopping/restarting, raising/lowering rate, hooking/unhooking, pump alarms, frequent gravity boluses, changing [the gastrostomy] tube site dressing, cleaning up formula spills...

Care provision as a *psychological stressor* (e.g., anxiety around central line infections and associated hospitalizations, distress related to the child's experience of pain or discomfort during dressing or tube changes, and parent worries about stalled growth) was also highlighted in 10 parent responses:

My child's failed growth sometimes feels like a personal failure. I feel like I didn't do enough to make him like food... [it makes] you feel as a parent, you're not doing something right!

Sleep

Half of the respondents reported sleep interruptions as having a strong negative impact on their well-being. In open-ended responses, parents described waking to respond to pump alarms or tend to their child's nighttime

feeds or toileting. Some specifically described the consequences of chronic sleep deprivation on their ability to function during the day:

Prolonged, broken sleep just sucks the life out of you... and has affected our happiness and ability to be functional and engaged elsewhere in life (work, relationships, extra-curricular activities, and self-care).

Mental health

Over a third of respondents (37%) perceived a strong negative impact of their child's medical procedures or hospital stays on their well-being. Open-ended responses for this item almost exclusively described the psychological impact of these experiences:

She gets extremely upset having to have any medical procedures, and I have to literally hold her down for them to get done. It tears me apart inside and makes me feel like I'm betraying her and hurting her myself...

Half of the respondents reported social isolation as having a strong negative impact; 40% perceived a strong negative impact stemming from the constant need to be prepared for an emergency to happen:

I feel that I'm frequently in a state of hypervigilance and basically afraid of the next bomb to drop. This causes me to feel anxious, irritable, and exhausted.

Worrying about their child was rated as having a strong negative impact on parent well-being more frequently (65%) than any other item on the survey. Open-ended responses to the follow-up questions for these items described the mental health ramifications of the child's illness journey on parents' overall well-being, including anxiety, stress, and depression. Some respondents described living in a near constant state of worry about what their child's future holds. Parent worries centered around their child's health in the future and their ability to live a "normal life" (eg, attend college, develop healthy peer relationships, care for themselves as an adult):

I always have somewhere in my mind thoughts/worries about his future and how he will manage on his own—how will he go to college, find a partner, etc. The worry is just always in my mind.

TABLE 3 Thematic analysis of open-ended parent responses about disease-specific impacts on parent quality of life ($n = 20$).

Disease-specific survey items	Subtheme (n of respondents)	Exemplary quotes selected from parent responses to open-ended follow-up questions
Provision of care		
Management of medical devices/therapies	Logistical stressors ($n = 10$)	“The task of preparing ([PN] in the past and) G-tube feedings every night is a burden.” “It can be exhausting keeping up with all the ordering, storing, organizing, and trialing new/different supplies—you feel like a case manager, pharmacist, and wound care nurse... among other titles.” “Potty training is difficult due to [PN]. He pees frequently and dealing with his line, backpack, and ostomy bag is hard to handle over a toilet in a small bathroom.” “The incredible amount of laundry generated by her is overwhelming. I’ve worked out a cleaning system over the last few years that helps get the stains out, but it’s still time-consuming and just plain gross.”
PN and EN management, central line care, feeding tube care/ostomy care, and child’s toileting and feeding	Psychological stressors ($n = 10$)	“She experiences pain with stooling and screams in pain. It is so, so hard to watch and not be able to do anything to help... sometimes suppositories are needed, and that is excruciating for me to watch or even hear being done from another room because she gets absolutely hysterical and fights and screams in pain.” “So many tantrums and tears over not being allowed to eat. [Our] relationship has significantly changed because I follow [the doctors’] orders for food”
Sleep		
Sleep interruptions	Poor parent sleep because of child’s nighttime waking or care needs ($n = 8$)	“Our son is actually a good sleeper, but his fear of medical stuff has spilled over into a fear of bugs in his room. We have had a few months of disrupted sleep. He also has to be changed a few times a night. My husband and I haven’t had a full night’s sleep in about 7 years.” “[My child] wakes twice a night for pooping, food, [and] drink. Never get a full night’s sleep”
	Consequences of chronic sleep deprivation ($n = 6$)	“When she was younger, we didn’t have overnight nursing (through our state’s Medicaid waiver for medically fragile kids), and sleep deprivation and depression/anxiety nearly put me in the hospital and nearly cost my husband his job.” “Not having energy to do simple tasks due to exhaustion”
Mental health		
Worries about future, worries about peers	Worries about child’s health ($n = 6$)	“We’re... fearful about how much enteral feeding will be tolerated, how long [PN] will be needed, and long-term impacts on health. We’re afraid of additional problems coming up, eg needing

(Continues)

TABLE 3 (Continued)

Disease-specific survey items	Subtheme (<i>n</i> of respondents)	Exemplary quotes selected from parent responses to open-ended follow-up questions
		<p>more bowel resected, line infections, liver problems, etc.” “One of my biggest fears is her having to go back on [PN], although I realize that most people with as severe SBS as she has, do end up back on it from time to time throughout their lives. But, I’m also afraid of the reality that any illness at all can put her life in jeopardy.”</p>
	<p>Worries about child’s ability to live a “normal” life in future (<i>n</i> = 7)</p>	<p>“His education: will he be able to get into college... and maintain the rigorous pace, along with some limitations in strength and vitality? Potential health issues (being out/missing class): will he be able to work a regular job and maintain employment (when he’s hospitalized, it can be 7 days to a few weeks). Not all employers are forgiving.” “I worry that he has so much... happening in his childhood that it will be impossible for him to have a happy, content life as an adult. I constantly worry about his future.”</p>
<p>Worries, always preparing for emergency, other negative impacts^a</p>	<p>General mental health including anxiety, hypervigilance, depression, and stress (<i>n</i> = 6)</p>	<p>“Always on high alert [of] something going wrong is awful. Constant stress, anxiety, fear, no time to be happy. [Too] scared to be happy [in case] something goes wrong” “I definitely operate under a ‘worst case scenario’ mindset all of the time.”</p>
<p>Isolation</p>	<p>Lack of connection or sense of shared experience; feelings of isolation (even prior to COVID-19) (<i>n</i> = 7)</p>	<p>“It’s been so isolating. I feel so isolated and alone. So often, we have had people: friends, family, and even medical staff who just ‘don’t get it.’”</p>
<p>Child’s hospitalizations, child’s medical procedures</p>	<p>Psychological burden (<i>n</i> = 6)</p>	<p>“Because she is immune suppressed due to missing a majority of her intestines and because even a cold puts her at risk for dehydration and hospitalization, we’ve always had to socially isolate to some extent... COVID-19 [has] added an additional layer of isolation that is now year-round.”</p>
		<p>“I have PTSD, depression, and anxiety from my son’s diagnoses and associated procedures. I always promised myself I would NEVER not be with him for a hard procedure, but having to hold him down and hear him say things and look at me like I am not protecting him and/or [like I’m a] part of the torture has been hard to deal with. I think it would have been beneficial for me to not always have been the one to restrain and hold him down; but alternatively, I also never wanted him to feel like I left him or wasn’t there! Handing your child over multiple times to the unknown and sitting and waiting for [the] hours-long surgery is not what you sign up for as a parent and it sucks.”</p>

TABLE 3 (Continued)

Disease-specific survey items	Subtheme (<i>n</i> of respondents)	Exemplary quotes selected from parent responses to open-ended follow-up questions
Access to resources and support Financial problems, lack of access to childcare or home nursing care, and other negative impacts ^a	Difficulty accessing trusted childcare or home nursing care (<i>n</i> = 7)	“There is NO childcare, respite, or down time. We are on 24/7 without much down time in between.” “I wouldn’t have ever let her be with anyone other than a nurse so that we could feel confident to go out. Since she’s been older and can assume a greater role in her line safety, we let her just be at [friends’] houses with the parents knowing she has her emergency kit and knows what to do.”
	Parent ability to work affected because of child’s complex needs (<i>n</i> = 6)	“Because of our daughter’s medical fragility and intense care needs and therapies, I haven’t been able to work since she was born... we struggle to make it on just my husband’s income, but between our daughter’s care needs and homeschooling, it’s just not feasible for me to work.” “Having to quit several jobs I loved due to [my child’s] lengthy hospitalizations or illnesses has been hard, and sad.”

Abbreviations: EN, enteral nutrition; G-tube, gastrostomy tube; PN, parenteral nutrition; PTSD, posttraumatic stress disorder; SBS, short bowel syndrome.

^aAdditional negative impacts included in subtheme were mentioned by parents in an open-ended follow-up question asking, “Aside from the items mentioned above, are there any other items that you would say have a negative impact on your overall well-being (ie, your overall sense of being comfortable, healthy, and happy)?”

Access to resources and support

Financial issues (including health insurance coverage) were reported by a quarter of respondents. Of the parents endorsing the lack of access to childcare and/or home nursing care items, 47% and 60%, respectively, perceived these items to have strong negative impact on their well-being. In open-ended responses, parents described the implications of their child’s condition for their ability to work; others described, rarely, if ever, getting breaks or opportunities to participate in restorative activities. Nearly half mentioned they had no one they trust and rely on to care for their child’s complex needs outside of the immediate family:

I am one of two people in the face of [the] earth that knows how she needs to be cared for and how she likes her dressing done and her ostomy done and how to vent her. [It] is very daunting.

Parent coping

When asked to report items that positively impact their well-being or ability to cope, 16 parents provided an

answer. Most commonly, parent responses centered the child, with parents describing the life lessons their child has taught them, their gratitude for their child’s survival, and the joy they experience because of their child:

[My son] is an awesome, resilient, basically healthy little boy. He makes my life richer despite his issues.

When I watch him grow/develop and exhibit joy and love, I have a great sense of relief and appreciation because the future is so uncertain.

Parents also described members of their support systems, which could include family, close friends, and trusted clinicians:

Finding a great [medical] team and the ensuing trust we have in them has been a huge positive part of this journey... that trust takes away some of the hopelessness and helplessness we feel.

For two parents, support received from and given to other parents through social media support groups has helped them cope:

Being a part of social media support groups and pages [related to SBS] has been helpful. You can learn a lot... but most importantly, you just don't feel so alone.

DISCUSSION

Children with SBS have complex care needs, most of which are met in the home setting by family caregivers who may experience stressors and strains unique to their role. Recent research indicates that parents of children with IF receiving PN spend nearly 30 h per week caring for their child's diverse condition-related needs.¹¹ Other studies present evidence of comparatively poorer HRQOL among families of children with SBS or IF, hypothesizing that the high level of care required by the child negatively impacts parent and family well-being.^{3,6,12} The results of the current study suggest that, although daily care is a stressor for parents, the negative impact of a child's SBS on parent well-being may stem largely from three overarching and interconnected themes: chronic sleep deprivation and its associated consequences, parent mental health, and difficulty accessing resources and support. Figure 2 outlines the complex and interrelated factors shaping well-being for parents of children with SBS, and offers strategies to assess and address them based in part on recommendations developed by the National Alliance for Caregiving.¹³

Provision of care

In the current study, some aspects of the day-to-day care for their child's SBS were perceived by parents to impact their own well-being negatively. Importantly, this negative impact did not exclusively stem from the logistical challenges associated with the child's condition management, but also from psychological challenges. Notably, although a child's PN dependence has been proposed as a likely driver of poor QoL for parents,^{6,12} PN management and central line care were among the least frequently reported negative impact items by survey respondents. Open-ended responses suggest that fear of infections and parents' hypervigilance associated with the child's central line, rather than the logistical management of PN, negatively impact parent well-being. As highlighted in other research,^{7,14} these findings suggest that the psychological stressors associated with caring for a child with SBS may be an important area of further research, assessment, and intervention.

Sleep

Half of parent respondents reported sleep interruptions as strongly negatively impacting their overall well-being. Interrupted and poor quality sleep has been previously reported as a problem for parents of children receiving PN^{3,15,16} or EN¹⁷ and for parents of children with chronic illnesses more generally.^{18,19} Because sleep is linked to mental and physical health,²⁰ memory,²¹ and daytime functioning,¹⁸ chronic sleep deprivation may impact parents of children with SBS in multiple well-being domains, including physical, emotional, social, and cognitive functioning. Although poorer HRQOL for parents of children with IF has been reported on these domains previously, the current study suggests sleep interruptions and associated fatigue may be a *driver* of these outcomes. This is another important area requiring additional research and intervention, because poor sleep may not only negatively shape a parent's well-being but may also impact the quality of care they are able to provide for their child.

Mental health

Issues related to parents' mental health, including frequent worrying, hypervigilance, anxiety, and trauma related to their child's medical journey repeatedly arose as themes across all survey domains. Even when reporting on the negative impact of items related to care provision, half of the qualitative responses described psychological stressors (e.g., anxiety surrounding possible central line infections and feelings of guilt due to stalled weight gain).

When asked directly about worries related to their child's health and future, parents reported this as a negative-impact item more frequently than any other item on the survey, a finding that aligns with previous research,^{3,6,12} and warrants special consideration. Children with SBS today are members of the first generation of SBS patients benefiting from advances in the field that have largely transformed the condition from a fatal disease to a chronic condition.¹⁻³ Still, parents searching for information after their child's SBS/IF diagnosis report difficulty accessing updated, reliable information that offers hope for their child's future.²² To help address parent worries, clinicians and researchers in the SBS/IF space should prioritize the effective and accessible communication of new research findings and current treatment paradigms through mechanisms such as community education webinars, open-access research articles, and publications in more general audience outlets.

Another set of mental health-related items warranting special discussion is hospitalizations and medical

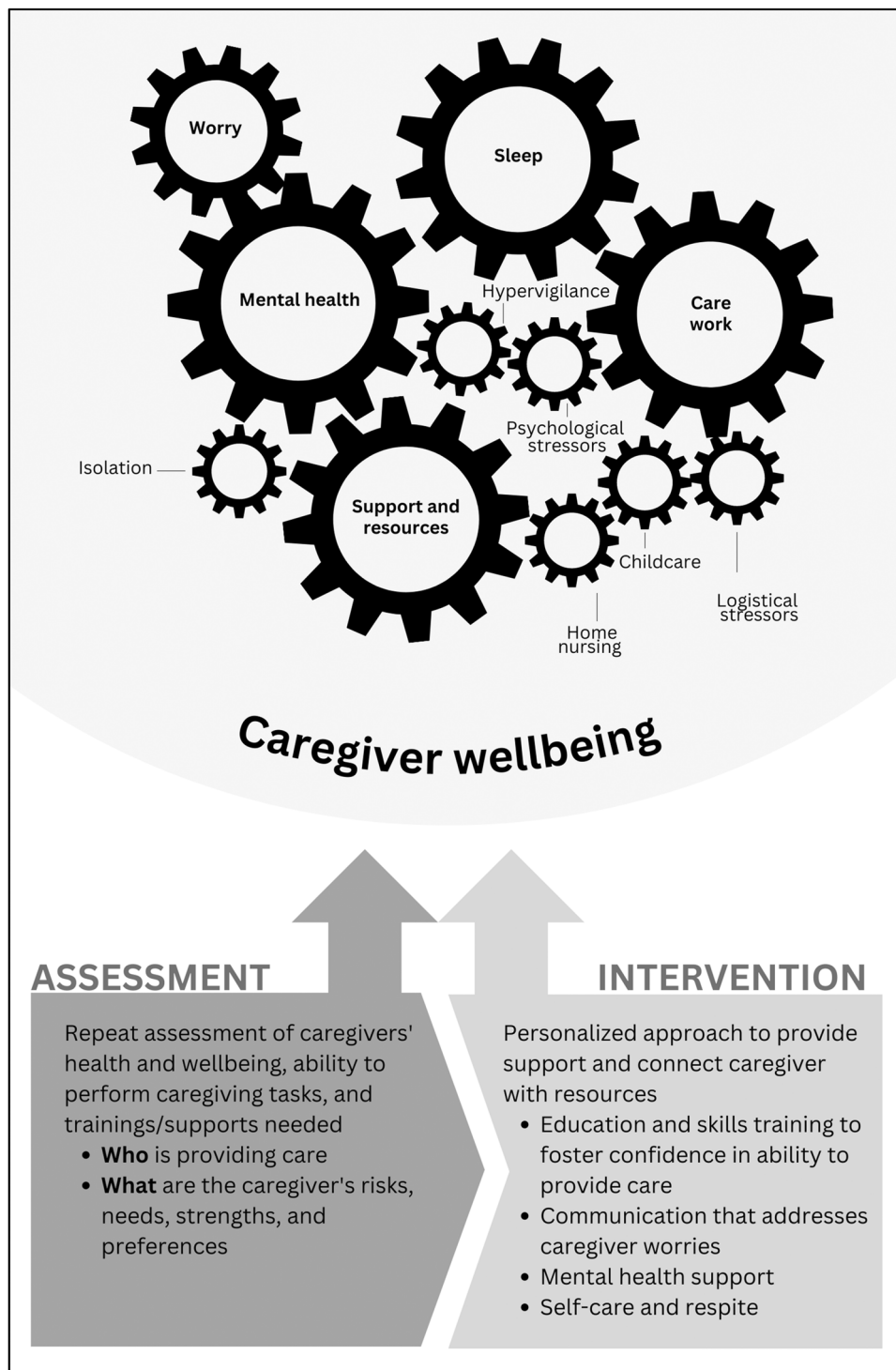


FIGURE 2 Conceptual model of the complex and interrelated factors shaping well-being for caregivers of a child with short bowel syndrome, and suggestions for assessing and addressing them. Suggestions are based in part on recommendations developed by the National Alliance for Caregiving as highlighted in “From Insight to Advocacy: Addressing Family Caregiving as a National Public Health Issue.”¹³

procedures. Although these two items were not originally developed to fit within the mental health category of the survey tool, qualitative analyses revealed the perceived negative impact of these two items stemmed almost exclusively from the psychological strain they cause

rather than from their logistical burden on parents (e.g., coordinating appointments). Some parents explicitly described the distress they felt because of their child’s multiple medical procedures or the trauma they experienced from restraining their child during medical

procedures. An emerging literature on pediatric medical trauma stress (PMTS) has estimated that roughly a fifth of children experiencing illness or injuries and their parents experience PMTS from their medical encounters.²³ For pediatric SBS patients and their families, the experience of medical trauma and its potential impact on QoL has received little attention in the literature and should be explored further in future research. It may also be an important area of intervention.

Access to resources and support

Because of a sense of social isolation (in part due to the COVID-19 pandemic) and a lack of access to trusted and high-quality care for their child, many parents reported feeling alone in managing of their child's complex care needs. Research has reported poorer social functioning for parents of children with SBS or IF^{6,12} and suggested a lack of support outside of the family as a potential barrier experienced by parents.²⁴ Although a lack of access to nursing care was not a problem uniformly across all survey respondents, 60% of the parents who reported this item as applicable perceived a lack of access to home nursing care as having a strong negative impact on their own well-being. Research has found a significant gap in the number of allotted versus received nursing hours for families of children with complex medical needs²⁵; this unmet need translates to missed employment, lack of sleep and opportunities for respite, and high levels of stress for parents,^{25,26} and drives hospital readmissions for children dependent on medical technology.²⁷ Without access to trusted support, whether in the shape of trained home care nurses, childcare providers, or members of personal support networks, parents must shoulder their child's care on their own. Better understanding the resource and support needs of parents to children with SBS is an important step to meeting those needs and improving outcomes.

Parent coping

Although understanding individual stressors associated with caring for a child with SBS offers valuable insights into possible interventions and future areas of research, it is important to recognize the caregiving experience as complex and multidimensional.⁸ Paralleling other research,^{3,8} parents in the current sample described the gratitude and joy they experience because of their child, and supportive relationships they have developed with others, including family, friends, clinicians. Highlighting parent and family resilience and strength while working to

address unmet needs adds important nuance to what could otherwise be understood as a burden-focused narrative.

Limitations

There are several limitations associated with the study sample and the survey tool. Most notably, the size and homogeneity of the study sample limit the generalizability of these findings. The sampling strategy, which relied on a convenience sample of members of a stakeholder committee, resulted in a survey sample that lacked in diversity, with respondents almost exclusively identifying as highly formally-educated, non-Hispanic White women from the Christian tradition. Further, the study sample likely underrepresented families with limited financial resources or language barriers to seeking care who might experience greater logistical challenges. The cross-sectional study design offers only a snapshot of the complex stressors and coping strategies that shape well-being for parents of children with SBS, and conducting this study during the COVID-19 pandemic may have additionally impacted the study results. Longitudinal data from a more diverse sample are needed to better understand these relationships.

CONCLUSION

Parents of children with SBS may face numerous stressors (and joys) that are unique to their caregiving role and may have implications for their health and their child's health and well-being. Our findings suggest that the logistical management of a child's medical therapies and SBS-related symptoms may be only partially responsible for the negative impact of a child's SBS on parent well-being. Chronic sleep deprivation, psychological stressors and parent mental health, and access to support and resources may play a critical role in shaping parent well-being outcomes. These findings suggest that unmet parent needs extend far beyond the clinical realm, highlighting possible avenues for future research and the development of targeted interventions to support caregivers.

Utilizing a family-centered approach to care, multi-disciplinary care teams should consider regular assessment of caregiver well-being to identify unmet needs and possible areas of intervention. The inclusion of mental health and/or palliative care providers as valued members of the interdisciplinary care team may help to ensure caregivers are supported and receive referrals to care and services when warranted. Ultimately, assessing and addressing the needs of family caregivers is an important part of caring for pediatric patients with SBS.

AUTHOR CONTRIBUTIONS

Marie L. Neumann, Jessica Y. Allen, Amy Ladner, Swapna R. Kakani, Meaghann S. Weaver, and David F. Mercer contributed to the conception and design of the research; Marie L. Neumann contributed to the acquisition, analysis, and interpretation of the data, Jessica Y. Allen and Amy Ladner contributed to the analysis and interpretation of the data; Marie L. Neumann drafted the manuscript, and all authors critically revised the manuscript. All authors agree to be fully accountable for ensuring the integrity and accuracy of the work and read and approved the final manuscript.

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CONFLICT OF INTEREST STATEMENT

Swapna Kakani is a speaker and has received consultant fees from Takeda and VectivBio AG. David F. Mercer received consultant fees from Zealand Pharma and Takeda, and is a primary investigator in trials sponsored by Zealand Pharma, VectivBio, and Takeda. All authors have no conflict of interest to declare in preparing this article. Meaghann S. Weaver contributed to this paper in a private capacity. No official support or endorsement by the US Department of Veterans Affairs is intended, nor should be inferred. Zealand Pharma, the study sponsor, had no role in the study design, data collection, analysis or interpretation, the writing of this manuscript, or the decision to submit this manuscript for publication. The first draft of the manuscript was written by Marie L. Neumann, and no honorarium, grant, or other form of payment was provided for the production of the manuscript.

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