



# “We Are Anxious Every Day”: COVID-19 Impacts on Youth with Spina Bifida

Colleen Stiles-Shields <sup>1</sup> PhD, Tessa K. Kritikos <sup>2</sup> PhD, Monique M. Ridosh,<sup>3</sup> PhD, Meredith Starnes,<sup>2</sup> BS, and Grayson N. Holmbeck,<sup>2</sup> PhD

<sup>1</sup>Section of Community Behavioral Health, Department of Psychiatry and Behavioral Sciences, Rush University Medical Center, Chicago, IL, USA, <sup>2</sup>Psychology Department, Loyola University Chicago, Chicago, IL, USA, and <sup>3</sup>Loyola University Chicago, Marcella Niehoff School of Nursing, Maywood, IL, USA

All correspondence concerning this article should be addressed to Colleen Stiles-Shields, PhD, Section of Community Behavioral Health, Department of Psychiatry and Behavioral Sciences, Rush University Medical Center, 1645 W. Jackson Blvd., Suite 302, Chicago, IL 60612, USA. E-mail: elizabeth\_stiles-shields@rush.edu

Received 23 February 2021; revisions received 27 May 2021; accepted 28 May 2021

## Abstract

**Objective** This study examined the impact of the COVID-19 pandemic on a national sample of adolescents and young adults (AYA) with spina bifida (SB) and parents of youth with SB. **Methods** AYA with SB (15–25;  $n = 298$ ) and parents of children with SB ( $n = 200$ ) were recruited to complete an anonymous, online survey in English or Spanish. Participants provided information about demographic and condition characteristics, as well as their technology access and use for behavioral health care. They also completed the COVID-19 Exposure and Family Impact Survey (CEFIS), which includes Exposure, Impact, and Distress subscales. Exploratory correlations and  $t$ -tests were used to examine potential associations between CEFIS scores and demographic, medical, and access characteristics. Qualitative data from the CEFIS were analyzed using thematic analysis. **Results** Scores on the Exposure, Impact, and Distress subscales demonstrated significant variability. Demographic associations with Exposure differed for those with higher Impact and Distress (e.g., White, non-Hispanic/Latino AYA reported higher rates of exposure [ $p = .001$ ]; AYA who identified with a minoritized racial/ethnic identity reported greater impact [ $p \leq .03$ ]). Impacts to mental and behavioral health ( $n = 44$ ), interference with medical care ( $n = 28$ ), and interpersonal challenges ( $n = 27$ ) were the most commonly occurring qualitative themes. **Conclusions** The current findings implicate differential impacts to individuals with SB and their families based on demographic, medical, and systemic factors (e.g., minoritized status). Recommendations to support families with SB and other pediatric conditions are made.

**Key words:** adolescents; COVID-19; emerging/young adults; health disparities and inequities; spina bifida.

## Introduction

The COVID-19 pandemic poses unique challenges to adolescents, young adults, and parents of children (under age 18). While nearly all individuals are currently struggling with fear, uncertainty, isolation, contact restrictions, and economic concerns, pediatric patients

are confronted with additional burdens. These include dramatic changes to their education, social connections, home life, and ability to seek medical care during a time of normative developmental milestones and medical autonomy development. Indeed, pediatric patients are living through a pandemic in the midst of

a developmental period where they are likely to have less well-developed emotional and cognitive resources while simultaneously managing these milestones. Moreover, their caregivers face increased responsibilities and demands of juggling parenting, work, home life, and ensuring adequate medical care and adherence with severely reduced support systems. Recognition of the cumulative stress of these demands on these pediatric patients and parents has led to a nascent literature exploring the impact of COVID-19 on pediatric patients and their parents and the creation of instruments such as the COVID-19 Exposure and Family Impact Survey (CEFIS; Kazak et al., 2021) to assess the impact of the pandemic on families.

Emerging evidence suggests that youth experience pandemics and quarantines differently than adults due to normative developmental differences. While the ways different age groups experience this pandemic may be unprecedented for all, the psychological impact of quarantine and stressors associated with the pandemic (e.g., prolonged duration, fears of infection, frustration and boredom, and decreased social connection) can be more severe and enduring for youth (Brooks et al., 2020; Wang et al., 2020). The manifestation of psychological distress in children also varies by developmental age. Younger children (3–6 years old) seem to exhibit clinginess and fear of infection, while older children (6–18 years old) more frequently demonstrate inattention and persistent questioning about the COVID-19 virus (Viner et al., 2020). Children's confusion and distress may be exacerbated by possible limitations in the areas of cognitive development and the comprehension of complex information about COVID-19, as well as the general uncertainty and lack of information surrounding the virus (Dalton et al., 2020; Provenzi et al., 2020). Children with special education needs may be particularly vulnerable to disruptions in routine, lack of structure, and suspension of important therapy and support services that are needed for their developmental progress (Lee, 2020).

Parents are also generally shouldering a greater burden than they did prepandemic with regards to childcare, education, and financial concerns (Fong & Iarocci, 2020; Marchetti et al., 2020). This increased burden can lead to feelings of burnout, exhaustion, and emotional distress. A national survey of parents of children under 18 years old in the United States found that since March 2020, 27% of parents reported worsening mental health for themselves and 14% reported worsening behavioral health for their children. Additionally, parents experienced food insecurity, decreased employer-sponsored insurance coverage, and loss of childcare (Patrick et al., 2020). Parenting a child with special needs has been identified as one predictor of greater parenting-related exhaustion in the

context of COVID-19 (Marchetti et al., 2020). The lack of support, reduced access to healthcare, therapies, and other support services, and concern about the safety of a vulnerable and/or immunocompromised child may explain this association.

Youth with spina bifida (SB) represent a population that is particularly vulnerable to the ramifications of the COVID-19 pandemic. SB is a congenital birth defect resulting from a failed closure of the embryonic neural tube (Copp et al., 2015). Adolescents and young adults with SB (AYA-SB; 15–25 years of age) are in a time of transition that can feel tumultuous, even in typical circumstances. They must negotiate a complex medical and self-care regimen, a range of significant comorbidities, as well as the normative developmental demands of adolescence. In the context of the COVID-19 pandemic, AYA-SB must also confront new barriers to adherence and self-management (Plevinsky et al., 2020), disrupted social relationships, increased time at home (which may be associated with increased family conflict), and greater increases in time accessing technology and social media (Psihogios et al., 2020). Variable cognitive and executive functioning deficits present in SB may also make it difficult to comprehend incoming information about the virus, reasons for social distancing, and quarantining (Stern et al., 2018). Parents of youth with SB (<18 years of age) already confront a range of challenges related to managing their child's medical regimen, concern about their child's health, and uncertainty regarding their child's independence (Mullins et al., 2007). Increased personal stress and parenting stress, which are heightened for parents in the context of the pandemic (Hiraoka & Tomoda, 2020), also predict greater parental perceptions of child vulnerability (Driscoll et al., 2018). This increased perception of child vulnerability may exacerbate worries about infection from the virus and ramifications of isolation, quarantine, and social distancing for their children with SB.

The current study aimed to examine the impact of the COVID-19 pandemic on a national sample of both AYA-SB (15–25 years) and parents of youth under the age of 18 with SB. The analyses were exploratory in nature but were hypothesized to identify needs specific to the vulnerable and often underserved population of youth with SB and their families. Specifically, Exposure, Impact, and Distress from the pandemic (Kazak et al., 2021) were explored for associations with (a) demographic and SB characteristics to identify disparities that may have occurred within this population; and (b) remote access options (e.g., Home Broadband/Wifi), given their importance for maintaining social distancing procedures for activities such as distance learning, telehealth appointments, etc. Examining the impact of the COVID-19 pandemic on

AYA-SB and parents of children with SB is critical to inform ongoing clinical care and support for youth with SB and may inform support for broader pediatric chronic health populations, including those with intellectual and physical disability components (Stiles-Shields et al., 2019).

## Methods

### Participants

The Spina Bifida Association (SBA), a national voluntary health organization supporting education, advocacy, and research to improve the lives of individuals with SB, advertised the study link on their website and social media platforms. Participants were eligible for inclusion if they were either an: (a) AYA with SB (15–25 years of age) and were able to answer the survey questions without the assistance of others (to ensure privacy and honesty in responses); or (b) parent of a child ( $\leq 18$  years of age) who has SB. Participants who both completed the survey and provided their email address (see Procedure section) were compensated for their time and participation with a \$5 Amazon Gift Code.

### Procedure

In compliance with Loyola University's Institutional Review Board, participants were consented via digital waiver of documented consent before participating. As such, interested participants clicked on a link displayed on recruitment materials that connected them to a detailed digital version of the study consent form. Participants were unable to proceed to the survey without first agreeing to participate by checking a "yes" box. Participants were then able to complete the survey after completing the digital waiver of documented consent. To maintain anonymity with survey responses, participants clicked on a separate link at the end of the survey that launched a new survey where they could provide their email address for compensation (i.e., participant email address was not able to be tied to their survey responses). Recruitment and consenting materials were provided in English and Spanish.

### Measures

Self-report assessments were administered and managed via the secure online survey platform, Qualtrics (Qualtrics, 2013), licensed and administered by Loyola University Chicago. All participants were able to complete the survey in English or Spanish.

### Demographic, Medical, and Access Characteristics

Participants were asked to report the following information: age, sex, race/ethnicity, education/employment status, and SB characteristics, including type of

SB, shunt status, shunt revision history, and lesion level. Consistent with variable selection for previous online surveys designed for AYA with SB (Stiles-Shields & Holmbeck, 2020), these SB characteristics were chosen to balance questionnaire brevity with relevant condition-specific variables for which both AYA with SB and parents of children with SB were likely to be able to provide valid reports (e.g., it was unrealistic to anticipate that participants would be able to report intelligence quotient scores). Participants were also asked to report: (a) whether they have home WiFi/Broadband Internet access; (b) if they sought mental or physical health care via technological platforms since March 2020; and (c) what type of visit/platform was used for such care. All demographic, medical, and access characteristic items were translated into Spanish by a native-speaking clinician (M.M.R.).

### COVID-19 Exposure and Family Impact Scale

The COVID-19 Exposure and Family Impact Scale (CEFIS; English and Spanish versions) is a brief self-report measure assessing the impact of the COVID-19 pandemic on families of youth with health conditions (Kazak et al., 2021). The CEFIS yields: (a) an Exposure score, which is calculated by summing 25 "yes/no" items assessing the exposure of families to COVID-19 related events (e.g., school closure, family member hospitalized for COVID-19); (b) an Impact score, which is calculated by averaging 10 items assessing the impact of COVID-19 on a family based on a four-point Likert scale (e.g., impacts to physical well-being, ease of parenting; an Impact score was not calculated for participants with more than three missing or "Not Applicable" responses); (c) a Distress score, which is calculated by averaging two items assessing the distress of parents and children on a 10-point Likert scale; and (d) an open-ended question to promote the sharing of details not covered in the previous sections. Higher scores across the Exposure and Distress scores indicated greater intensity across these domains. As the Impact score is averaged from a four-point Likert scale, a positive valence interpretation (i.e., less impact) occurs for scores  $\leq 2.5$  and a negative valence (i.e., more impact) interpretation occurs for scores  $> 2.5$ . The mean Exposure score for the initial validation of the CEFIS was  $8.71 \pm 4.07$  (range 0–25); the mean Impact score for the CEFIS was  $2.68 \pm 0.65$  (range 1–4); and mean caregiver self-reported parental and child Distress scores were  $5.95 \pm 2.34$  and  $5.44 \pm 2.55$  (range 1–10), respectively (Kazak et al., 2021). The CEFIS Exposure ( $\alpha = .77$ ), Impact ( $\alpha = .87$ ), and family-level Distress subscales ( $\alpha = .68$ ) demonstrated acceptable reliability for the current sample.

The COVID-19 Exposure and Family Impact Scale, Adolescent and Young Adult Version (CEFIS-AYA;

English and Spanish versions) yields similar scores to the CEFIS, with the following exceptions: (a) the Exposure score consists of an additional three items so that specific experiences could be queried for both the AYA and a family member (e.g., “I had to...”; “A member of the family had to...”); (b) the Impact score consists of an additional six items to assess domains specific to the psychosocial development of AYA (independence, sedentary behavior, substance abuse, loneliness, friends, and dating); and (c) the Distress score is comprised of only one item (i.e., rating of personal distress only). Findings from the open-ended question of the CEFIS-AYA were not reported in the current study due to the low frequency of responses (i.e., <20%). While normative data and clinical cut-offs for the CEFIS-AYA are anticipated as forthcoming from the creators, they are not available at this time. The CEFIS-AYA Exposure ( $\alpha = .89$ ) and Impact subscales ( $\alpha = .86$ ; the Distress subscale score is comprised of only one item for AYA and therefore is not reported) demonstrated acceptable reliability for the current sample.

### Data Analysis

Descriptive analyses were used to characterize the sample and the CEFIS Exposure, Impact, and Distress subscale scores. Exploratory correlations and *t*-tests were used to examine potential associations between CEFIS and CEFIS-AYA scores with demographic, medical, and access characteristics. Variables with low cell counts (i.e., <40; education, employment, home Broadband/Wifi, and smartphone app use) were not analyzed for potential associations (e.g., only nine parents who reported not having Home Broadband Internet/Wifi had complete CEFIS subscale scores). Narratives from the parent-reported, open-ended question were analyzed using thematic analysis. Specifically, the first and second authors both coded the responses and discussed any conflicts in the rating until a consensus was reached. Data were coded using an inductive approach, such that decisions were driven by the data, rather than theory (Patton, 1990).

## Results

### Sample Characteristics

Participants were recruited in September and October of 2020. Three hundred and forty AYA participants (15–25 years) and 228 parent participants completed consent for the study. Following the removal of participants with incomplete or inaccurate data (see [Supplementary Table 1](#) for data exclusion details), 298 AYA and 200 parent participants were included in the current study. The AYA-SB participants were divided fairly evenly by gender (51% male). Most of the sample identified as White, Non-Hispanic/Latino

(64.4%), and often had the most severe type of SB (myelomeningocele; 41.6%), with a shunt (60.1%) and a lumbar lesion level (54.4%). AYA participants ranged in age from 15 to 25, with an average age of 21. Sixty-three percent of parent participants identified as female and about three-quarters identified as White, Non-Hispanic/Latino. Their children with SB had an average age of 10.12 (SD = 4.98) and were more typically male (57%) with myelomeningocele (56.5%), a shunt (53.5%), and a lumbar lesion level (66%). [Table 1](#) displays the sample demographics, medical, and technology access characteristics.

### Impact of the COVID-19 Pandemic

#### Adolescents and Young Adults with SB

AYA-SB-reported Exposure, Impact, and Distress were significantly correlated with one another, such that higher Exposure was associated with lower Impact ( $r = -.18, p = .02$ ) and higher Distress ( $r = .31, p < .001$ ). Impact and Distress were positively correlated ( $r = .16, p = .01$ ). AYA-SB reported an average Exposure score of  $7.62 \pm 5.97$  (range 0–25). Higher Exposure scores were associated with a higher number of shunt revisions ( $r = .42, p < .001$ ; see [Supplementary Table 2](#)). AYA-SB who identified as White, non-Hispanic/Latino, and those who had myelomeningocele reported higher exposure ( $p = .001, d = .79$ ). Higher exposure was also associated with seeking behavioral health treatment via technology ( $p < .001, d = 1.73$ ). Further, those who reported having a video visit with their physician ( $p < .001, d = .84$ ) or having a phone visit with their physician reported higher exposure ( $p < .001, d = 1.21$ ).

The average Impact score was positively valenced (i.e., < 2.5; less impact) at  $2.41 \pm .52$  (range 1–4). Higher Impact scores were associated with having a higher number of shunt revisions ( $r = .24, p = .03$ ). Impact was also associated with race/ethnicity, such that AYA who identified with a minoritized racial or ethnic identity reported higher impact ( $p = .03, d = .30$ ). Higher Impact was also associated with having a video ( $p < .001, d = .50$ ) or not having a telephone visit with a physician ( $p = .009, d = .34$ ). The average Distress score suggested a moderate experience of distress ( $5.85 \pm 2.06$ ; range 1–10). Higher AYA Distress was associated with a higher number of shunt revisions ( $r = .31, p = .001$ ). Distress was also higher for those who sought treatment via technology ( $p < .001, d = .52$ ). Higher distress was also associated with having a video ( $p = .004, d = .36$ ) or phone visit with a physician ( $p < .001, d = .42$ ).

#### Parents of Children with SB

*Exposure, Impact, Distress.* For parents of youth with SB (>18 years), there was no evidence to suggest associations among the CEFIS subscales, with the exception

**Table I.** Demographic, Medical, and Access Characteristics

	AYA ( <i>n</i> = 298)	Parent ( <i>n</i> = 200)
Age	21.35 (2.20)	38.16 (6.94)
Parent report of child's age	–	10.12 (4.98)
Gender		
Male	152 (51.0%)	73 (36.5%)
Female	145 (48.7%)	127 (63.5%)
Other <sup>a</sup>	1 (0.3%)	–
Parent report of child's gender		
Male	–	114 (57.0%)
Female	–	86 (43.0%)
Race, ethnicity		
African American/Black	46 (15.4%)	16 (8.0%)
Asian	22 (7.0%)	2 (1.0%)
White, Hispanic/Latino	38 (12.8%)	26 (13.0%)
White, Non-Hispanic/Latino	192 (64.4%)	155 (77.5%)
More than one race	1 (0.3%)	1 (0.5%)
Highest level of education		
Finished grade school	3 (1%)	1 (.5%)
Some high school	49 (16.4%)	5 (2.5%)
Finished high school	82 (27.5%)	17 (8.5%)
Some college	89 (29.9%)	59 (29.0%)
Finished college	25 (8.4%)	77 (38.5%)
Business or technical school	47 (15.8%)	14 (7.0%)
Attended graduate school or professional school after college	3 (1.0%)	6 (3.0%)
Received a professional degree	–	22 (11.0%)
Employment status		
Student	182 (61.1%)	3 (1.5%)
Employed, full time	23 (7.7%)	128 (64.0%)
Employed, part time	29 (9.7%)	21 (10.5%)
On disability for work	14 (4.7%)	3 (1.5%)
Unemployed	47 (15.8%)	42 (21.0%)
Volunteer	3 (1.0%)	3 (1.5%)
SB type		
Myelomeningocele	124 (41.6%)	113 (56.5%)
Meningocele	48 (16.1%)	23 (11.5%)
Lipomeningocele	51 (17.1%)	26 (13.0%)
Lipomyelomeningocele	37 (12.4%)	21 (10.5%)
Lipoma	34 (11.4%)	14 (7.0%)
Unsure	4 (1.3%)	3 (1.5%)
Shunt	179 (60.1%)	107 (53.5%)
Shunt revision	146 (81.56%)	62 (57.9%)
Number of shunt revisions	2.27 (2.78)	2.38 (1.91)
Lesion level		
Sacral	56 (18.8%)	38 (17.5%)
Lumbar	162 (54.4%)	132 (66.0%)
Thoracic	68 (22.8%)	26 (13.0%)
Unsure	12 (4.0%)	7 (3.5%)
Has home broadband/WiFi	280 (94.3%)	191 (95.5%)
Access use or changes since March 2020		
Sought care via technology (generally)	159 (53.4%)	107 (53.5%)
Had video visit with physician	89 (29.9%)	80 (40%)
Had telephone visit with physician	104 (34.9%)	51 (25.5%)
Used smartphone/tablet apps to monitor physical health	53 (17.8%)	16 (8.0%)
Used smartphone/tablet apps to monitor mental health	75 (25.2%)	17 (8.5%)
Used other Technology for Behavioral Health Management	–	4 (2.0%)

Note. SB = spina bifida.

<sup>a</sup>The participant who selected “Other” for their gender did not further specify with what gender identity they identify.

of higher Exposure being associated with greater Distress ( $r = .17, p = .02$ ). This stands in contrast to the initial psychometric findings of the CEFIS, which identified a low magnitude, but significant association of Exposure and Impact ( $r = .11, p < .0001$ ); and a

significant association for Distress with both Exposure and Impact (Kazak et al., 2021). Parents of children with SB reported an average Exposure score of  $8.65 \pm 3.83$  (range 0–21), which is comparable to scores collected from caregivers of multiple pediatric



populations ( $8.71 \pm 4.07$ ; Kazak et al., 2021). Exposure was negatively correlated with parent age ( $r = -.16$ ,  $p = .03$ ; see [Supplementary Table 3](#)). Exposure was also associated with their child's gender ( $p = .009$ ,  $d = .39$ ), such that parents of females reported higher exposure. Parents who reported changing their home internet access since the onset of the pandemic ( $p < .001$ ,  $d = .75$ ) and who sought treatment via technology ( $p = .002$ ,  $d = .46$ ) and who reported completing a video visit with their child's physician reported higher levels of exposure ( $p = .001$ ,  $d = .49$ ).

Parental Impact was negatively valenced (i.e.,  $>2.5$ ; more impact), such that the average score was  $2.65 \pm .60$  (range 1–3.8). Again, this is a comparable finding to the Impact scores yielded from caregivers of multiple pediatric populations ( $2.68 \pm 0.65$ ; Kazak et al., 2021). [Supplementary Figure 1](#) displays the means for individual Impact items, with all Personal Well-being items demonstrating more impact ( $>2.5$ ): Anxiety (3.03), Mood (2.84), Exercise (2.73), Eating (2.71), and Sleeping (2.67), as well as select Family Interactions (Parenting [2.59], Caring for Older Adults/Others in Family with Disabilities [2.54], and Caring for Child with a Medical Condition [2.51]). Impact was associated with parental gender and parental race/ethnicity, such that mothers ( $p = .02$ ,  $d = .38$ ) and parents who identified as White, Non-Hispanic/Latino reported higher impact ( $p < .001$ ,  $d = .65$ ). To better understand this difference of impact experienced based on racial or ethnic identity, the Impact scale factors of Personal Well-Being (items 30–35) and Family Interactions (items 26–30) were examined. However, there was no evidence to suggest differences in impact based on racial or ethnic identity across these two factors (Personal Well-Being,  $t = -1.13$ ,  $p = .3$ ; Family Interactions,  $t = -1.12$ ,  $p = .3$ ). Finally, the average family Distress score was moderate, at  $6.28 \pm 1.42$  (range 2–8.5). This subscale score was broken down into personal distress level ( $6.47 \pm 1.56$ ; range 2–9) and parent-report of their child's level of distress ( $6.02 \pm 1.81$ ; range 2–9). Family Distress was associated with parental gender, such that fathers reported higher levels of family distress ( $p = .009$ ,  $d = .40$ ). Distress was also associated with parental race/ethnicity, such that parents who identified as White, non-Hispanic/Latino reported higher family distress ( $p = .01$ ,  $d = .50$ ).

*Other COVID-19 Effects on Families with a Child with SB: Open-Ended Responses.* One hundred and twenty-six parent participants (55.3%) provided responses on the open-ended item regarding other negative and/or positive effects of the pandemic on their children and families. The most commonly occurring themes centered on impacts to mental and behavioral health ( $n = 44$ ),

interference with medical care ( $n = 28$ ), and interpersonal challenges ( $n = 27$ ). Most commonly, parents described impacts on the daily mental and behavioral health of their children and families (e.g., “We are anxious every day.”). For those with younger children with SB, difficulties in processing the need for reduced social contact led to an increase in behavioral and mental health challenges:

Since our SB daughter is too young to understand what was happening and why we couldn't go see people or go to gymboree or play group, she started acting out more and became more easily aggravated. . . It's been a very difficult time for her to adjust from going out and socializing all the time to being stuck in the house. She has become more independent when it comes to learning how to play and entertain herself, which is great, but lock down has definitely taken a toll on her mental health since she is a social butterfly and so used to getting out of the house multiple times a week to participate in classes and see people.

Many parents also detailed the need to assume extra responsibilities to manage their child's SB in the face of less accessible medical care (e.g., “As a parent, it is extremely difficult to be a speech therapist, physical therapist, occupational therapist, wound care nurse, vision therapist, and a general Ed teacher on top of being a parent.”). Further, some parents noted that decreased medical care has led to declines in their child's functioning, such that “. . . Greatly reduced [occupational therapy] OT and [physical therapy] PT, delayed surgery and inability to get therapies led to loss of mobility-related skills and confidence that are ongoing.” In addition to impacts on decreased medical care, many parents noted the interpersonal difficulties surrounding isolating from family and friends (e.g., “COVID-19 has affected us as far as not being able to see other family members, we are close knit so it's a struggle not being able to see them.”). Relatedly, many parents noted frustration in maintaining social distancing as other families appear to not be doing the same. For example, “Another big source of sadness is that we see so many other people going back to normal and not following guidelines for groups or masks. It seems like everyone else is returning to normal while we sit on the sidelines, still quarantined. That is the hardest thing to explain to the kids.”

Other themes that emerged from the data included interference with recreational activities ( $n = 18$ ; “He also cannot take his swim lessons or ride his horse. It has not been good for his muscles.”), financial and economic concerns ( $n = 18$ ; “The financial pressure is huge.”), school/education ( $n = 16$ ; “Putting a 3 year old in front of a screen to learn special Ed is unacceptable and not sustainable.”), and generic negative impacts ( $n = 11$ ; “Our lives have become more intense.”). Fear of infection and/or a concern regarding having a child that is immunocompromised also emerged as a theme ( $n = 6$ ; “I'm afraid of getting infected by going out to work every day.”). While the

majority of comments centered on negative impacts, positive impacts were noted by 26 parents (e.g., “Learned a lot more about what my son is able to accomplish with a little more help.”).

## Discussion

The current study examined the impact of the COVID-19 pandemic on a national sample of AYA-SB (15–25 years) and parents of youth with SB (<18 years). The use of the CEFIS to conduct an initial assessment about the impact of the pandemic on these samples was based on an examination of both quantitative data and open-ended question responses. Given the diverse symptom and self-management profiles of youth with SB (Stiles-Shields et al., 2020) and the myriad accompanying challenges that these profiles pose for youth and their parents, these families provide an excellent model for studying the impact of COVID-19. While our aims were exploratory, we expected that the COVID-19 pandemic and its associated features (e.g., frustration and boredom, lack of social connection) would negatively impact these families. Further, we intended to use the findings to inform recommendations for moving forward during the pandemic, not only for families of youth with SB but also for other families with chronic pediatric conditions.

Given that (a) this study used recruitment methods designed to reach a national sample via an online survey; and (b) this exploratory study is the first to examine the impact of the COVID-19 pandemic on families of individuals with SB, we were cautious not to over-interpret findings. However, these results suggest several interesting conclusions regarding the impact of this pandemic on this group. In terms of demographic and SB characteristics, the following factors were associated with higher exposure to COVID-19: higher number of shunt revisions (AYA-reported); identifying as White, non-Hispanic/Latino (AYA); having myelomeningocele (AYA); parent being younger in age (parent-reported); and having a female child with SB (parent). While some associations may be circumstantial (e.g., more parent participants had children who were female than male), having a higher number of shunt revisions or having myelomeningocele (both implicated in greater SB severity) may indicate that higher treatment needs left some families at greater risk for exposure to the virus (e.g., receiving necessary care in-person despite social distancing recommendations). If this were the case, it is possible that these greater needs led to changes in technology use and access over time. Indeed, Exposure was also associated with telehealth and technology access and use, such that higher exposure was endorsed by those who reported: seeking treatment via technology (AYA, parent); having a video visit with their physician (AYA,

parent); and having a phone visit with their physician (AYA). Of note, the associations for higher exposure differed from those with higher impact and distress. Specifically, higher impact scores were associated with: identifying with a minoritized racial or ethnic identity (AYA); higher number of shunt revisions (AYA); being female (parent); and having a video or telephone visit with a physician (AYA). Distress was associated with a higher number of shunt revisions (AYA); being male (parent); identifying as White, Non-Hispanic (parent); seeking treatment via technology (AYA); and having a video or telephone visit with a physician (AYA).

The differential impact of the COVID-19 pandemic on minoritized groups (Gemelas et al., 2021; Smith & Judd, 2020), an experience that is co-occurring with a “racism pandemic” (Valenzuela et al., 2020), is reflected in the current findings. For example, although White, Non-Hispanic/Latino youth had higher self-reported rates of exposure, it was youth who identified with a minoritized racial or ethnic identity that experienced the greater self-reported impact of the pandemic on their lives. Sensitivity analyses to explore whether this impact may have been driven by family interactions or personal well-being were inconclusive. However, there is a growing literature identifying that the pandemic has had greater impacts on minoritized groups (Gemelas et al., 2021; Smith & Judd, 2020; including the level of care that is received, Mulchan et al., 2021).

The short- and long-term mental health impacts from this pandemic are anticipated to be significant for adults and children alike (Galea et al., 2020). The feedback from parents reflected this notion, as much of the impact of the pandemic on families has centered on mental and behavioral health challenges. Further, the isolation from others required by social distancing has limited their children’s abilities to socialize, attend school, and receive their typical medical care. Such limitations have resulted in behavioral problems, frustration, declines in children’s physical functioning (e.g., mobility-related skills impacted by a reduction in occupational and physical therapies), and the need for parents to assume multiple roles (e.g., parent, teacher, physical therapist). Families with a child with multiple shunt revisions may be especially at risk for such burdens. Indeed, higher numbers of shunt revisions were associated with greater AYA-reported exposure, impact, and distress. Having a shunt is associated with cognitive correlates (Hommeyer et al., 1999), which may be one of the multiple factors that increase the difficulties associated with the COVID-19 pandemic for such AYA-SB. Further, each shunt revision necessitates major brain surgery; AYA-SB may be fearful that the virus would require more hospitalizations and/or traumatic medical experiences beyond what they have

already experienced. These findings highlight both (a) the impact of the COVID-19 pandemic on AYA-SB and their families; and (b) the need for further research on the ways in which this pandemic has differentially affected different groups of youth with SB (e.g., those with shunt revisions, those with more severe forms of SB, and those with fewer sociodemographic resources).

### Recommendations

Parents reported difficulties managing mental and behavioral changes in their children, as well as concerns about their medical care, education, and social disruptions. Further, frustration was voiced in seeing other populations “return to normal” while youth with SB and their families continue to social distance. Given the need to continue to socially distance while dealing with the frustrations of doing so, telehealth and digital mental health tools (e.g., COVID Coach, SuperBetter; [Psihogios et al., 2020](#)) may be beneficial for both youth with SB and parents to find support and increase exposure to evidence-based coping skills for these frustrations. In response to the pandemic and moving forward, the integration of pediatric psychologists in healthcare systems serving families of youth with SB would also promote early interventions, mental health supports, and the potential to promote means to address implicit biases that exacerbate pediatric health disparities ([Mulchan et al., 2021](#)). To further support mental health, the Spina Bifida Association (SBA) recommends that families try to connect with others safely, control their intake of COVID-19-related news, stay physically active, and maintain a routine as much as possible ([Brei & Dicianno, 2020](#)). In terms of concerns about youth with SB falling behind in medical and SB-related care due to the pandemic, the Spina Bifida Association (SBA) has made recommendations for families to use this time at home while social distancing to promote developmental milestones and self-management skills for children with SB. Specifically: (a) focus on parent self-care activities to promote well-being for parents and children with SB alike (youth with SB are predisposed to depression, anxiety, and other mental health concerns; [Kritikos et al., 2020](#)); (b) teach and practice skills around self-management and independence skills (e.g., self-catheterization, self-bowel management); (c) fine-tune bowel and bladder management programs; (d) attend telehealth visits as much as possible (as opposed to attending in-person); (e) continue to utilize and communicate with school administrators regarding Individualized Educational Plans/504 plans, which remain in force and which schools are required to maintain through e-learning; and (f) continue to address transitioning from pediatric to adult healthcare ([Brei & Dicianno, 2020](#)).

### Implications for Other Pediatric Populations

Given the complex nature of SB and its overlapping symptom profiles with other pediatric conditions, these findings and recommendations extend to other pediatric populations and their parents as well. Indeed, immunocompromised youth, such as AYA with cancer, may also confront elevated anxiety ([Košir et al., 2020](#)), fears of contamination and prolonged isolation, and social distancing, beyond what their healthy counterparts are experiencing. Medically complex youth, such as those with cystic fibrosis, may experience similar challenges navigating and attending multisystem health care appointments and various needed services, potentially impacting their disease management and treatment adherence ([Plevinsky et al., 2020](#)). Additionally, youth with neurocognitive differences, such as those with Attention-Deficit/Hyperactivity Disorder or Intellectual Disability, face similar barriers with regards to remote learning and its negative impact on their education and specialized services ([Becker et al., 2020](#)). Thus, the recommendations included here may be extended to these youth and their parents as well. Indeed, it is essential that families of youth with pediatric illnesses be empowered during this pandemic and beyond with resources, support and advocacy, education, and monitoring for mental health and psychological well-being ([Serlachius et al., 2020](#)). Further, other pediatric specialties would also benefit from the integration of pediatric psychologists to promote early intervention and additional supports for families.

### Limitations and Future Directions

The current study had multiple strengths, including the use of a measure specific to the experience of AYA and parents of children with a pediatric condition ([Kazak et al., 2021](#)), recruitment methods designed to reach a large national sample, and timely findings on the experience of families during an unprecedented, international pandemic. However, there are important limitations to consider in interpreting these findings. Indeed, as location was not assessed, the national distribution of this sample cannot be demonstrated. These data were also collected at a single time point about 9 months into the pandemic (United States) and situated at the start of an academic year. It is unclear how the findings generalize to experiences at other points in the pandemic or across the pandemic. Additionally, while online surveys have previously been demonstrated as feasible for AYA-SB ([Stiles-Shields & Holmbeck, 2020](#)), there are inherent limitations to this methodology, such as misinterpretation of questions without the ability to directly clarify the meaning of specific items with research personnel. Further, the need to answer questions independently may have led to a sample of AYA with less disease



severity. The use of only virtual recruitment and data collection methodologies likely also excluded the participation of youth and parents with limited or no internet access. Even in the case of online surveys, future research should ensure a more equitable recruitment approach (e.g., provide tablets in clinics to complete the survey in waiting rooms). Further, equitable recruitment and data collection methodologies could be better informed through the input of a stakeholder advisory board to ensure that the research practices best meet the lived experience of AYA-SB and parents of youth with SB (Stiles-Shields et al., 2020). Such research practices are more likely to promote diverse and representative sampling, increasing the ability of future research to identify differential predictors of needs and outcomes to best support targeted interventions and advocacy.

The current study employed recruitment and study materials in both English and Spanish, yet the sample was primarily White, non-Hispanic/Latino (64.4% for AYA, 77.5% for parents). As SB has the highest occurrence in Hispanic populations (i.e., 3.8 for every 10,000 live births), as compared to non-Hispanic White (3.09/10,000) and non-Hispanic Black or African American populations (2.73/10,000; Canfield et al., 2014), it was anticipated that the use of a bilingual methodology would improve the representative nature of this sample compared to previous online surveys with people with SB. However, none of the surveys were completed in Spanish. While it was anticipated that AYA-SB may be more likely to engage with the survey in English, the parent data did not demonstrate any preference for the study materials in Spanish. One reason may be that the parent sample was relatively young ( $M$  age = 38.16), as compared to a previous in-person, longitudinal sample of Hispanic/Latino adults (Gallo et al., 2014). Yet, another potential reason may be that providing study materials in Spanish in isolation is an inadequate engagement method. As such, recommendations for future research endeavors to better foster engagement with Hispanic/Latino populations include: (a) creating a “personal touch” through direct contact with researchers (García et al., 2017); (b) engaging and nurturing community connections; (c) employing potential participants in recruitment roles and compensating them for their time; and (d) harnessing the social networks of community members (Rhodes et al., 2018).

## Conclusion

To the best of our knowledge, the current study is the first to examine the impact of COVID-19 on families with a child with SB. The findings of this study characterize the impact of this pandemic on the lives of AYA-SB and the parents of youth with SB and reveal

the disparate ways in which this impact may affect specific groups (e.g., those with more shunt revisions). It is recommended that families with youth with SB are recommended to maintain self-care, focus on SB self-management, and use telehealth and other digital tools for care and social connection. Further, inclusive research methodologies are warranted to continue to better understand the impacts of the pandemic on families of children with a pediatric condition.

## Supplementary Data

Supplementary data can be found at: <https://academic.oup.com/jpepsy>.

## Funding

This research was supported in part by a grant from the National Institute of Nursing Research and the Office of Behavioral and Social Sciences Research (R01 NR016235; K01 NR018907). Dr. Stiles-Shields is also supported by a fellowship from the Cohn Family Foundation and an R3 funding mechanism supported by Rush University Medical Center and RTI International.

## Acknowledgments

The authors thank the Spina Bifida Association (SBA), without whom this research would not have been possible. Correspondence regarding this manuscript can be sent to [gholmbe@luc.edu](mailto:gholmbe@luc.edu).

*Conflicts of interest:* None declared.

## References

- Brooks, S. K., Webster, R. K., Smith, L. E., Woodland, L., Wessely, S., Greenberg, N., & Rubin, G. J. (2020). The psychological impact of quarantine and how to reduce it: rapid review of the evidence. *The Lancet*, *395*, 912–920.
- Canfield, M. A., Mai, C. T., Wang, Y., O'Halloran, A., Marengo, L. K., Olney, R. S., Borger, C. L., Rutkowski, R., Fornoff, J., Irwin, N., Copeland, G., Flood, T. J., Meyer, R. E., Rickard, R., Alverson, C. J., Sweatlock, J., & Kirby, R. S.; for the National Birth Defects Prevention Network (2014). The association between race/ethnicity and major birth defects in the United States, 1999–2007. *American Journal of Public Health*, *104*(9), e14–e23.
- Copp, A. J., Adzick, N. S., Chitty, L. S., Fletcher, J. M., Holmbeck, G. N., & Shaw, G. M. (2015). Spina bifida. *Nature Reviews. Disease Primers*, *1*, 15007.
- Dalton, L., Rapa, E., & Stein, A. (2020). Protecting the psychological health of children through effective communication about COVID-19. *The Lancet. Child & Adolescent Health*, *4*(5), 346–347.
- Driscoll, C. F. B., Stern, A., Ohanian, D., Fernandes, N., Crowe, A. N., Ahmed, S. S., & Holmbeck, G. N. (2018). Parental perceptions of child vulnerability in families of youth with spina bifida: the role of parental distress and parenting stress. *Journal of Pediatric Psychology*, *43*(5), 513–524.

- Fong, V. C., & Iarocci, G. (2020). Child and family outcomes following pandemics: a systematic review and recommendations on COVID-19 policies. *Journal of Pediatric Psychology, 45*(10), 1124–1143.
- Galea, S., Merchant, R. M., & Lurie, N. (2020). The mental health consequences of COVID-19 and physical distancing: the need for prevention and early intervention. *JAMA Internal Medicine, 180*(6), 817.
- Gallo, L. C., Penedo, F. J., Carnethon, M., Isasi, C. R., Sotres-Alvarez, D., Malcarne, V. L., Roesch, S. C., Youngblood, M. E., Daviglius, M. L., Gonzalez, P., & Talavera, G. T. (2014). The Hispanic Community Health Study/Study of Latinos Sociocultural Ancillary Study: sample, design, and procedures. *Ethnicity & Disease, 24*(1), 77–83.
- García, A. A., Zuñiga, J. A., & Lagon, C. (2017). A personal touch: the most important strategy for recruiting Latino research participants. *Journal of Transcultural Nursing: Official Journal of the Transcultural Nursing Society, 28*(4), 342–347.
- Gemelas, J., Davison, J., Keltner, C., & Ing, S. (2021). Inequities in employment by race, ethnicity, and sector during COVID-19. *Journal of Racial and Ethnic Health Disparities*. 10.1007/s40615-021-00963-3.
- Hiraoka, D., & Tomoda, A. (2020). Relationship between parenting stress and school closures due to the COVID-19 pandemic. *Psychiatry and Clinical Neurosciences, 74*(9), 497–498.
- Hommeyer, J. S., Holmbeck, G. N., Wills, K. E., & Coers, S. (1999). Condition severity and psychosocial functioning in pre-adolescents with spina bifida: disentangling proximal functional status and distal adjustment outcomes. *Journal of Pediatric Psychology, 24*(6), 499–509.
- Kazak, A. E., Alderfer, M., Enlow, P. T., Lewis, A. M., Vega, G., Barakat, L., Kassam-Adams, N., Pai, A., Canter, K. S., Hildenbrand, A. K., McDonnell, G. A., Price, J., Schultz, C., Sood, E., & Phan, T.-L. (2021). COVID-19 exposure and family impact scales: factor structure and initial psychometrics. *Journal of Pediatric Psychology, jsab026*. 10.1093/jpepsy/jsab026.
- Košir, U., Loades, M., Wild, J., Wiedemann, M., Krajnc, A., Roškar, S., & Bowes, L. (2020). The impact of COVID-19 on the cancer care of adolescents and young adults and their well-being: results from an online survey conducted in the early stages of the pandemic. *Cancer, 126*(19), 4414–4422.
- Kritikos, T. K., Smith, K., & Holmbeck, G. N. (2020). Mental health guidelines for the care of people with spina bifida. *Journal of Pediatric Rehabilitation Medicine, 13*(4), 525–534.
- Lee, J. (2020). Mental health effects of school closures during COVID-19. *The Lancet. Child & Adolescent Health, 4*(6), 421.
- Marchetti, D., Fontanesi, L., Mazza, C., Di Giandomenico, S., Roma, P., & Verrocchio, M. C. (2020). Parenting-related exhaustion during the Italian COVID-19 lockdown. *Journal of Pediatric Psychology, 45*(10), 1114–1123.
- Mulchan, S. S., Wakefield, E. O., & Santos, M. (2021). What COVID-19 teaches us about implicit bias in pediatric health care. *Journal of Pediatric Psychology, 46*(2), 138–143.
- Mullins, L. L., Wolfe-Christensen, C., Pai, A. L. H., Carpentier, M. Y., Gillaspay, S., Cheek, J., & Page, M. (2007). The relationship of parental overprotection, perceived child vulnerability, and parenting stress to uncertainty in youth with chronic illness. *Journal of Pediatric Psychology, 32*(8), 973–982.
- Patrick, S. W., Henkhaus, L. E., Zickafoose, J. S., Lovell, K., Halvorson, A., Loch, S., Letterie, M., & Davis, M. M. (2020). Well-being of parents and children during the COVID-19 pandemic: a national survey. *Pediatrics, 146*(4), e2020016824.
- Patton, M. Q. (1990). *Qualitative evaluation and research methods* (2nd ed. 532). Sage Productions, Inc.
- Plevinsky, J. M., Young, M. A., Carmody, J. K., Durkin, L. K., Gamwell, K. L., Klages, K. L., Ghosh, S., & Hommel, K. A. (2020). The impact of COVID-19 on pediatric adherence and self-management. *Journal of Pediatric Psychology, 45*(9), 977–982.
- Provenzi, L., Baroffio, E., Ligabue, S., & Borgatti, R. (2020). The little professor and the virus: scaffolding children's meaning making during the COVID-19 emergency. *Frontiers in Psychiatry/Frontiers Research Foundation, 11*, 817.
- Psihogios, A. M., Stiles-Shields, C., & Neary, M. (2020). The needle in the haystack: identifying credible mobile health apps for pediatric populations during a pandemic and beyond. *Journal of Pediatric Psychology, 45*(10), 1106–1113. <https://doi.org/10.1093/jpepsy/jsaa094>
- Qualtrics, I. (2013). Qualtrics. Provo, UT, USA.
- Rhodes, S. D., Alonzo, J., Mann-Jackson, L., Tanner, A. E., Vissman, A. T., Martinez, O., Rodriguez-Celedon, R., Garcia, J. M., Arellano, J. E., Song, E. Y., Eng, E., & Reboussin, B. A. (2018). Selling the product: strategies to increase recruitment and retention of Spanish-speaking Latinos in biomedical research. *Journal of Clinical and Translational Science, 2*(3), 147–155.
- Serlachius, A., Badawy, S. M., & Thabrew, H. (2020). Psychosocial challenges and opportunities for youth with chronic health conditions during the COVID-19 pandemic. *JMIR Pediatrics and Parenting, 3*(2), e23057.
- Smith, J. A., & Judd, J. (2020). COVID-19: vulnerability and the power of privilege in a pandemic. *Health Promotion Journal of Australia, 31*(2), 158–160.
- Stern, A., Driscoll, C. F. B., Ohanian, D., & Holmbeck, G. N. (2018). A longitudinal study of depressive symptoms, neuropsychological functioning, and medical responsibility in youth with spina bifida: examining direct and mediating pathways. *Journal of Pediatric Psychology, 43*(8), 895–905.
- Stiles-Shields, C., & Holmbeck, G. N. (2020). Health behaviors and disordered eating in adolescents and young adults with spina bifida: results from a national survey. *Disability & Rehabilitation, 42*(20), 2910–2916.
- Stiles-Shields, C., Plevinsky, J. M., Psihogios, A. M., & Holmbeck, G. N. (2020). Considerations and future directions for conducting clinical research with pediatric populations during the COVID-19 pandemic. *Journal of Pediatric Psychology, 45*(7), 720–724.
- Valenzuela, J., Crosby, L. E., & Harrison, R. R. (2020). Commentary: reflections on the COVID-19 pandemic and

- health disparities in pediatric psychology. *Journal of Pediatric Psychology*, 45(8), 839–841.
- Viner, R. M., Russell, S. J., Croker, H., Packer, J., Ward, J., Stansfield, C., Mytton, O., Bonnell, C., & Booy, R. (2020). School closure and management practices during coronavirus outbreaks including COVID-19: a rapid systematic review. *Lancet Child & Adolescent Health*, 4(5), 397–404.
- Wang, G., Zhang, Y., Zhao, J., Zhang, J., & Jiang, F. (2020). Mitigate the effects of home confinement on children during the COVID-19 outbreak. *The Lancet*, 395, 945–947.