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Parents' report on the health care management of spina bifida in early childhood

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Abstract

PURPOSE: This study aimed to describe health care use by type of health providers and care settings visited by children with spina bifida (SB) and to compare this use between children with and without a shunt.

METHODS: Health care use data were extracted from a larger study on the health and functioning of children with SB aged 3–6 years. The present study focused on the medical information subsection of a parent-reported survey related to SB care, general care, specialty care (e.g., neurosurgery), emergency care, and complications related to SB and shunts.

RESULTS: Parents of 101 children with SB participated. Most of the children were male with myelomeningocele and had a shunt. They visited a health care provider for SB care an average of 7.4 times and a specialist an average of 11.9 times in the previous 12 months. Most visited a multidisciplinary clinic for SB-related care and a private physician's office for general care. Children with a shunt had more SB-related medical visits, more visits to a specialist, and a greater number of different types of specialists than those without it. Frequency of emergency room visits did not differ between the two groups. Health providers informed parents about headaches, vomiting, and fever as signs of complications, and some parents did report shunt-related complications.

CONCLUSIONS: SB is a complex medical condition requiring that children receive medical care from various medical specialists, especially for children with a shunt. Findings on health care use

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suggest high levels of monitoring and care coordination that parents navigate to care for their child.

Keywords

Spina bifida; health care management; myelomeningocele

1. Introduction

Spina bifida (SB) is a birth defect affecting the spine that may cause physical and cognitive disability, depending on the type of diagnosis and the presence of comorbidities [1]. The most common types of SB are myelomeningocele, meningocele, and lipomeningocele. Myelomeningocele, in addition to being the most common, is also the most physically disabling due to abnormalities of the spinal cord, meninges, and bones. Although all types of SB require long-term follow-up after surgery for their care, myelomeningocele is the most medically complicated. The focus of health care management for individuals with SB shifts with their developmental stage as they transition through life. In early childhood, individuals with myelomeningocele are likely to undergo surgical procedures like shunt placement or revision and spinal closure [2-4]. Additionally, typical developmental milestones, such as bowel/bladder training, could be more difficult for children with SB to achieve compared to their developing peers [5]. Other associated conditions, such as skin breakdowns, could arise as many children with SB depend on wheelchairs and orthoses to perform their activities as they grow into adolescence [6].

Children with SB may require visits to different specialists as they develop shunt complications, bowel/bladder incontinence, hip dislocation, scoliosis, and delayed motor development. Studies on health care utilization suggest that many specialists are involved in the management of children with SB: neurologists, neurosurgeons, urologists, vision specialists, occupational and physical therapists, nurses, orthopedists, and nutritionists [7]; however, there are limited data on the utilization by discipline. Because of the complexity of the disability and the many health care specialties involved in the comprehensive care of someone with SB, care coordination is beneficial. Often, parents and caregivers assume the role of care coordinator [8] and, therefore, it is helpful for them to understand the importance of managing the substantial demands brought by caring for a child with SB [9-11]. Prior research has found that children with SB tend to use medical services at much greater frequency than their typically developing peers [12]. Ouyang and colleagues [13] examined health insurance claims data for privately-insured children and adults with and without SB, using a database of employer-sponsored insurance claims data. They found that health expenditures of children with SB were 13 times greater than those without SB and that children between one and 17 years old had an average of 23 outpatient visits within a year. Although these findings confirmed that health care utilization is much greater for those with SB, the study did not report on the type of health care specialists visited or on the specific settings where the care took place, beyond inpatient or outpatient. Medical record systems across different providers are typically fragmented; therefore, the breadth and depth of health services needed to provide care for individuals with SB is difficult to ascertain. Part of the goal of the current study was to describe, with greater nuance, health care usage.

Understanding whom the family sees for care, where they go for care, and what possible complications could arise from SB could help caregivers and health providers anticipate, prepare, and access financial, social, and other supports needed to gain good care.

Cassell and colleagues' study of Medicare expenditures [3] provided specific insights on the medical disciplines likely to be consulted for the management of SB. Claims data for children continually enrolled in Medicaid in North Carolina between 1995 and 2002 indicated that developmental/behavioral and rehabilitative services expenditures for children with SB were 82 times higher for children with SB than children who did not have a major birth defect. The authors suggested that the remarkable difference in expenditures could be explained by the relatively infrequent use of mental and rehabilitative care services among typically developing children. Inpatient care accounted for most health expenditures regardless of SB status. Infants with SB had an average of 2.6 times higher expenditure than infants without SB. Findings confirmed that health care use among those with SB is disproportionately greater than among those without SB or any major birth defect as well as that having hydrocephalus is associated with even greater health care use and expenses. However, the study only gave insight on general areas of care and settings but did not specify the types of specialists and the frequency with which they were visited. Information about care by specialists is useful because it could be a crude indicator of the gaps in the professional management of the many health and medical issues of individuals with SB.

The purpose of this study was to describe the reported use of health care services among children with SB aged 3–6 years based on frequency of visits, types of specialists seen, and places of care. A broad yet specific description of health care use may provide insight on the level of coordination necessary to manage this complex condition. Secondary purposes were 1) to examine whether having a shunt was associated with the use of health care services, with the presumption that children with a shunt would have more health visits in a year than those without a shunt, 2) to explore how health care providers educated parents about complications related to SB, and 3) to assess the frequency of shunt complications.

2. Methods

This project was part of a larger study on the health status and social and cognitive development of children with SB aged 3–6 years. Two sites, Utah and Arizona, participated in the project. Recruitment and participation occurred between May 2011 and September 2013. The larger study identified 152 eligible children with SB whose parents were contacted for participation. Of those contacted, 101 agreed to participate. Participant data came from multiple sources, including standardized questionnaires about the child completed by parents; medical records; and parent-reported surveys on their family functioning, child's behaviors, child's self-care, child's mobility, and child's health and well-being. Some children underwent neuropsychological testing. Participants were given the option of participating in a full or partial protocol, as explained further below. Children's medical records were abstracted with parental consent. More specific information regarding recruitment and participation are explained in a previously published methods paper [14].

2.1. Participants

Inclusion criteria were children with a confirmed diagnosis of SB between three and six years old at enrollment whose parents spoke Spanish or English. Although participants were initially identified from surveillance systems in both sites, the recruitment differed between sites due to site-specific Institutional Review Board (IRB) requirements. Separate IRB approvals were obtained for the two sites. Utah recruited eligible participants from the Utah Birth Defect Network, a population-based surveillance program. Additionally, parents of eligible children with SB attending the SB clinic during the study period were invited to participate. This occasionally included families attending the SB clinic from Utah's surrounding states (e.g., Idaho, Wyoming, Nevada). Arizona reached participants through recruitment letters, emails, and web announcements about the study. In-person recruitment at a SB clinic was allowed for only one of two Arizona sites. Arizona could only recruit from parents who replied to an open call for participants.

2.2. Procedure

Parents consented to participate in either a partial or a full protocol. Partial protocols consisted of a phone survey and mailed questionnaires. The full protocol was an in-person clinic visit with the parents and child in which the parents completed questionnaires and the child was administered standardized neuropsychological assessments by a licensed psychologist. All parents were asked to consent for abstraction of their child's medical records in search of data about hospitalizations, surgeries, and comorbidities.

Six parent-reported standardized questionnaires consisted of various validated assessments of behavioral, functional, and quality of life characteristics. In addition, a parent-reported 120-item survey developed for this project assessed health care visits, general health, medical information, mobility and functioning, nutrition and physical growth, and family demographics in this population. Data presented here are from the medical information section of the parent-reported survey, in which questions focused on SB-related care, places of care, and specialist care. Parents were asked about health care services used in the previous 12 months and about their child's shunt, when applicable.

2.2.1. SB-related care—Parents were asked about the number of visits their child had for SB-related care: "During the past 12 months, how many times did 'X' see a doctor, nurse, or other health care professional to receive care or treatment directly related to spina bifida? Please do not include well visits or general physical exams." They were also asked about the most frequent place of care: "Where does 'X' receive his/her spina bifida related medical care most often?" Parents' perception of adequacy of care was assessed: "During the past 12 months, would you say that 'X' received all the spina bifida related care that he/she needed?"

2.2.2. General care—Parents were also asked about the frequency with which their child received wellness care: "During the past 12 months, how many times did 'X' see a doctor or nurse for general preventive care, such as a physical exam or a well-child check-up? Please do not include doctor/clinic visits specifically related to spina bifida (e.g.,

visits to a neurosurgeon or urologist).” as well as location of care: “Where does ‘X’ receive his/her general/preventive medical care most often?”

2.2.3. Specialist care—Parents were asked about whether their child visited a specialist in the past year: “During the past 12 months, has ‘X’ been seen by any specialists?” They were also asked about the specific type of specialist visited in the past year: “Tell me if ‘X’ has been seen by a urologist, orthopedist, neurologist, ophthalmologist, neurosurgeon, or another specialist. How many times in the past 12 months?” The total number of visits to all specialists was summed, and the number of different types of specialists visited was calculated for each child.

2.2.4. Emergency visits—Parents also reported about general emergency care that their child received: “During the past 12 months, how many times did ‘X’ go to the emergency room for any reason?” and about SB-related emergency care in particular: “Of those [emergency] visits, how many times were these visits related to the spina bifida?”

2.2.5. Health providers’ message about complications—Parents reported on their child’s shunt status and complications that their health care providers had told them to pay close attention to: “Infants and toddlers with spina bifida can experience additional complications that need medical care. I am going to read you a fist of items; please let me know whether or not any of your health care providers have told you to watch out for any of these symptoms in ‘X’.” These items were complaints of headaches, difficulties eating, gagging, weak cry, fussiness, arm weakness, high pitched cry, noisy breathing, cyanosis (turning blue), difficulties breathing, vomiting, fever, and other.

2.2.6. Shunt-related complications—Occurrences of shunt obstruction and blockage, infection, revision, and replacement were assessed for those who had a shunt: “Has the shunt ever been [obstructed or blocked, infected, revised, replaced]?” In addition, parents were asked about the number of times that each of the shunt complications occurred in the child’s life.

Statistical analyses

IBM Statistical Package for the Social Sciences (SPSS) Statistics, version 25, was used to perform statistical analyses [15]. Mann-Whitney U tests were used to evaluate differences between those with and without a shunt regarding the number of visits for care related to SB, wellness care, specialists, and the emergency room (ER). Pearson’s chi-square test of independence was used to assess the association between shunt status and visits (yes or no) to specific specialists: urologist, neurosurgeon, orthopedist, neurologist, and ophthalmologist.

3. Results

3.1. Parents’ and children’s characteristics

Characteristics of parent-respondents and their children are described in Table 1. Primary insurance at the time of the study was not available, but primary insurance at birth

was. However, primary insurance at birth for 56 (55%) children was missing; 27 (27%) had Medicaid, 16 (16%) had private insurance, one (1%) had self-pay, and one (1%) had federal insurance. Other demographic information is published elsewhere [16]. Because there was no difference in health care visits for care related to SB, general care, specialist care, and emergency between children with myelomeningocele and non-myelomeningocele (lipomeningocele and meningocele), the two SB types were merged for the analyses of health care use. However, since all the children with a shunt had myelomeningocele, subanalyses were conducted testing group differences according to shunt for the myelomeningocele group.

3.2. SB-related care

Parents reported that their child visited a health provider for SB-related care an average of 7.4 times (n = 97, range: 0–50, standard deviation (SD) = 8.9, Median (Md) = 5.0) in the prior 12 months. This number excluded well-child preventive or general care visits. Four parents (4%) had missing visits information. Overall, those with a shunt had significantly more visits than those without (Md = 6.0, mean (M) = 8.7, SD = 9.9 vs Md = 2.0, M = 4.4, SD = 5.0, U = 1350.5, p = 0.007). Among the subset with myelomeningocele, no statistically significant difference was found between those without a shunt (Md = 3.0, M = 4.5, SD = 4.4) and with a shunt (Md = 6.0, M = 8.7, SD = 9.9, U = 474.5, p = 0.070).

Most parents indicated that they most often received SB-related medical care at a multidisciplinary clinic that specialized in SB (Table 2). Regarding perception of adequacy of care, 86 (85.1%) respondents indicated that their child received all the SB-related care they needed. Eleven (10.9%) indicated that they were not receiving the SB care needed (don’t know/missing, n = 4, 4.0%).

3.3. General care

On average, parents reported that their child saw a health care provider 2.0 times in the prior 12 months for general preventive care (n = 97, SD = 2.3, Md = 1.0, range: 0–12). Four (4.0%) parents had missing responses. Most parents reported that their child most often visited a private physician’s office for general preventive care (Table 2). There was no difference in the average number of general preventive care visits between children who had a shunt (Md = 1.0, M = 2.2, SD = 2.5) and those who did not (Md = 1.0, M = 1.7, SD = 1.4, U = 1095.5, p = 0.349). Among children with myelomeningocele, no difference in the average number of general care visits by shunt status was found (U = 462.5, p = 0.422).

3.4. Specialists visits

Of 101 parents, 92 (91.1%) reported that the child had seen a specialist in the prior 12 months, six (6%) did not report having seen a specialist, and three (3%) had missing information. Those who did report having gone to a specialist visited a median of four different types of specialists (range: 0–8). Table 3 lists the types of specialists visited. Overall, among those who reported having seen a specialist, children with a shunt had seen a greater number of different types of specialists (Md = 4.5, M = 4.7, SD = 1.3) than those without (Md = 4.0, M = 4.0, SD = 1.7, U = 1076.0, p = 0.049). However, among the myelomeningocele subset, the number of different specialists visited did not differ between

those with a shunt (*Md* = 4.5, *M* = 4.7, *SD* = 1.3) and those without (*Md* = 4.0, *M* = 4.3, *SD* = 1.7, *U* = 466.0, *p* = 0.310).

For those who reported having visited a specialist across all SB types combined, the average number of visits was 11.3 (*SD* = 11.5, *Md* = 8). Overall, those with a shunt had a greater number of visits than those without (*Md* = 8.5, *M* = 12.9, *SD* = 12.7 vs *Md* = 5.0, *M* = 7.0, *SD* = 5.5, *U* = 1131.0, *p* = 0.006). For children with myelomeningocele type, those with a shunt were reported to have had a greater number of visits (*Md* = 8.5, *M* = 12.9, *SD* = 12.7) than those without (*Md* = 4.5, *M* = 5.4, *SD* = 3.7, *U* = 587.5, *p* = 0.008).

Pearson's Chi-square tests for independence between shunt status and visits to specialists were conducted for the most frequently visited specialists: urologist, neurosurgeon, orthopedist, neurologist, and ophthalmologist. Findings indicated significant associations between having a shunt and visits to a urologist (χ^2 (1, N = 92) = 7.87, *p* = 0.005), neurosurgeon (χ^2 (1, N = 92) = 6.79, *p* = 0.009), and orthopedist (χ^2 (1, N = 92) = 8.27, *p* = 0.004). Only three individuals did not visit a urologist and, because two cells had fewer than 5 observations, a Fisher's Exact test was also conducted and supported the association between urology visit and shunt (*p* = 0.021). No relationship was found between shunt status and visits to a neurologist (χ^2 (1, N = 92) = 0.744, *p* = 0.388) or an ophthalmologist (χ^2 (1, N = 92) = 0.000, *p* = 0.992).

3.5. Emergency visits

Fifty (50%) respondents indicated that their child visited the ER in the previous 12 months. Forty-eight (48%) did not go to the ER, and three (3%) had missing data. Of the 50 who had an ER visit, 31 (62%) indicated that at least one of the visits was related to SB. The average number of SB-related visits in the previous 12 months was one (*SD* = 1.4). There was no difference in the average number of ER visits between those who had a shunt (*Md* = 1.0, *M* = 1.1, *SD* = 1.6) compared to those without a shunt (*Md* = 0.0, *M* = 0.6, *SD* = 0.8, *U* = 1140.5, *p* = 0.315). A sub-analysis among children with myelomeningocele indicated no difference in the number of ER visits by shunt status (*U* = 474.5, *p* = 0.653).

3.6. Health providers' message about complications

Parents reported the SB-related complications of which their health provider made them aware. More than half of parents were warned about headaches, vomiting, fever, difficulties with eating, and fussiness as well as other symptoms (Table 4).

3.7. Shunt-related complications

Of the 68 children who had a shunt, the shunt had been obstructed or blocked in 31 (46%), infected in 12 (18%), revised in 29 (42%), and replaced in 28 (41%). On average, respondents indicated that in their child's lifetime the child's shunt had been obstructed or blocked 2.6 times (*n* = 31, *SD* = 2.6, *Md* = 1.0), infected 1.1 times (*n* = 12, *SD* = 0.3, *Md* = 1.0), revised 2.7 times (*n* = 29, *SD* = 3.9, *Md* = 1.0), and replaced 2.0 times (*n* = 28, *SD* = 2.7, *Md* = 1.0).

4. Discussion

This analysis reports on the use of health care services among children with SB aged 3–6 years old as reported by their parents. The children visited a doctor or other healthcare provider in the previous 12 months for SB-related care an average of 7.4 times, and these visits were most likely to take place at a multidisciplinary clinic. The average number of general care visits was two times a year and most likely to be at a private physician's office. The median number of visits to specialists in the previous 12 months was eight, on average 11.9 visits. The frequency of health provider visits found here was lower than Ouyang and colleagues' finding [13] that children with SB aged 1–17 made an average of 23 outpatient visits during the year. However, the current study was limited to children 3–6 years old and not directly comparable to the authors' assessment of privately-insured individuals across pediatric age groups. Additionally, parent-reported information in this study was subject to recall bias, which may have under- or over-estimated actual healthcare utilization, as opposed to Ouyang et al.'s study, which was based on claims data. Nonetheless, these results indicated that use of health services from specialists among young children with SB was high compared to the general population of children under six years, of whom only 7% had over 10 health visits in 2017 [17]. Health insurance information was not available at the time of the study; therefore, it was not possible to assess the degree to which insurance status influenced the frequency of health visits.

Findings indicated that shunt status could be related to usage of some categories of health care. The prediction that children with a shunt would have more frequent health care use was partially supported. Among those with myelomeningocele, those with a shunt did have a greater number of visits to specialists, although the average number of different types of specialists did not differ by shunt status. The difference in the number of specialists visited between those with and without a shunt was found even within the myelomeningocele group. This difference may be driven by the severity of myelomeningocele, and shunt status could be indicative of secondary complications requiring the attention of a specialist. Children with a shunt, on average, had a significantly greater number of SB-related health visits (to a doctor or other health professional). However, these effects may have been driven by five outliers who all had a shunt and very high frequency of visits at two standard deviations greater than the mean and by relatively low visits among the children with non-myelomeningocele who all had no shunt. The aforementioned outliers were not removed because they likely reflect a real extreme in the range of the number of visits of SB-related care that children with a shunt seek. These findings could indicate that, although most who have shunts receive SB-related care with a frequency similar to that of individuals without a shunt, some children with a shunt might need an extraordinarily high amount of care. However, this difference does not necessarily indicate that shunts cause greater illness but that there could be a greater number of complications preexisting among those who need a shunt. Thus, the effect of shunt on frequency of visits must be interpreted with caution in that other congenital anomalies or illness were not accounted for, which could explain the greater number of health visits among those with a shunt.

Contrary to prediction, there was no difference in the average number of wellness or general care visits between those with and without a shunt. Both groups had an overall

low frequency of visits to a primary care provider. One possible reason is that a visit to a specialist for specific care may supersede the need for a wellness visit. Another reason is that issues typically discussed with the primary care providers are likely addressed by the specialists. On average, there were more frequent visits to specialists than primary care or emergency department visits. Children with a shunt also saw a greater number of different types of specialists. On average, the children with SB saw four different types of specialists in the previous year. Having a shunt was associated with visits to a urologist, orthopedist, and neurosurgeon. It is likely that these three types of specialists were the most visited because genitourinary conditions due to urinary tract infections and neurological complications of hydrocephalus are common conditions for individuals with SB [18]. The high frequency of neurosurgical and orthopedic specialist visits could also be due to shunt-, hip-, and spine-related complications. It was somewhat unexpected, however, that children with shunts were not more likely to see a neurologist, but it is plausible that neurology-related concerns may have been addressed by neurosurgeons. Over 40% of the parents in this study whose child had a shunt indicated that the child's shunt had ever been obstructed, blocked, revised, or replaced. These complications are likely to prompt a visit to the neurosurgeon. Only four parents reported a visit to a psychologist. This is noteworthy because children with spina bifida, particularly those with a shunt, tend to score lower on some measures of cognitive functioning [19, 20]. Although not assessed in this study, one possible explanation is lack of access to qualified health providers for testing. Another possible explanation is that, in early childhood, cognitive delays may not be evident to caregivers until the child undergoes neuropsychological testing.

Although beyond the scope of this study, accounting for the cause of ER visits may be useful for understanding health issues that may be missed during planned healthcare visits. Future research might examine reasons for ER visits among young children with SB. Half of the parents reported that their child had an ER visit at least once in the previous 12 months; this was a high rate compared to the general population. Future research could examine the reasons for ER visits, which may provide greater understanding of health issues missed during planned healthcare visits. Estimates from the National Health Interview Survey, a household survey of the civilian noninstitutionalized population in the United States, indicated that 24.4% of children aged 0–4 years and 17.7% of those aged 5–11 years had at least one ER visit in the previous 12 months [21], much lower than the 50% found in this study. Shunt status was not related to ER visits. This finding is notable because acute neurological issues is one of the top reasons for visiting the emergency department among individuals with SB [22]. A possible reason for the finding that shunt status was not related to emergency department visits is that health providers may encourage parents of very young children with SB to be vigilant about signs and symptoms of shunt complications and follow them closely. Proactive care may protect against complications and, therefore, reduce likelihood of unplanned health visits. Urinary tract infections can also be very painful, and parents may opt to take their child to the ER for fast pain relief if no other clinics are open or available to see them on short notice. The current study's population could be unique in that they may have been less reliant on the emergency department for care. Youths tend to use the emergency department less than adults with SB [23], perhaps because adults have less access to non-emergency care. It is also possible that children are more likely to have

access to insurance or benefits that allow them to attend SB clinics more frequently than adults. Future research could examine the degree to which health insurance, its type and quality of coverage, affects the types of services used, quantity of use, and, ultimately, health outcomes.

Parents gave information about potential complications related to SB to watch for, according to their health provider, and shunt-related issues that their child might experience. The most common complications that health providers educated parents about were headache, vomiting, and fever. The focus on headache education may be due to the high proportion of children in the study who had shunts, as headaches are common signs of an obstructed shunt. Out of the fist of complications queried, health providers might have encouraged parents to pay attention to fever and vomiting more frequently because these could be indicators of infection.

For optimal health, individuals with SB, especially those with myelomeningocele with a shunt, need multidisciplinary care. The finding indicating that children visit several different specialists multiple times a year brings into question whether management and coordination is undertaken mostly by parents or by health coordinators as part of the child's health care team. If the former, future investigations might assess the effect of personal burden on parents in seeking and receiving care for their child, potential implications on the functioning of the family unit, and ways to address the negative impact of potential fiscal, psychological, and social burdens [10]. Burden on families could be reduced by creating a health system that follows an efficient model of care for people with complex medical conditions. For example, Kinsman and colleagues [7] have suggested a multidimensional, multidisciplinary care model that is comprehensive, coordinated, and longitudinal. A comprehensive model would address the needs of the patient and of the family. A coordinated one would consist of a team of professionals with a range of clinical expertise who work together and with the family to develop the patient's plan of care. A longitudinal model has a health care team that provides the patient and family anticipatory guidance as the needs of the patient shift and transition across the lifespan. The present study's findings indicated that, for care related to SB, families did visit multidisciplinary clinics that specialized in SB. Future investigations might examine the degree to which care at these multidisciplinary clinics is comprehensive and considerate of patients' transitions across developmental stages, such as transitioning from pediatric to adult care [24]. For example, more information is needed on how to efficiently handle transfer of health information through the transition from pediatric to adult care. Transitioning to adult care may mean greater independence from parents or caregivers in making health-related decisions [24]. Self-advocacy and health literacy become important skills for patients at this stage. Additionally, studies evaluating the link between multidisciplinary care and patient outcomes, such as the potential benefits of multidisciplinary care on the prevention of secondary conditions, are needed.

5. Limitations

These findings should be interpreted with caution because results may not represent health care use by all children with SB. Utah's sampling frame for recruitment came from a

population-based state birth defect registry and some recruitment occurred via SB clinics. Arizona identified and recruited from a variety of sources including multispecialty clinics and hospital databases. Because some recruitment was clinic-based, it is likely that the study captured individuals who were in most need of, sought, and received care at SB clinics. Indeed, over 83% of the children in this study had myelomeningocele, a serious and potentially disabling type of SB. In addition, based on the high degree of health care utilization, parent-respondents in this study could represent the most proactive, and as such, were more prone to participate in the study, more informed about care for children with SB, and, therefore, most likely to seek help. The data support this assumption in that, when asked about adequacy of care, a large majority, over 85%, indicated that their child received the SB-related health care they needed. Taken together, the findings on the frequency of health service use could be high estimates compared to the overall population of children with SB. The current study did not assess the degree of time and effort required in caring for a child with SB, though study findings could be an indication. Future investigations could consider time and effort in addition to frequency. Additionally, not accounted for in the current investigation are other services that may be necessary for the management of SB, such as vendors involved in procurement of mobility equipment and urologic and continence management supplies. Thus, findings presented here account for only some of the network of services that parents or caregivers may need to provide the recommended care for their child.

6. Conclusions

SB is a complex medical condition that requires multidisciplinary care. Findings about the frequency with which parents reported visits to a health care provider in the previous 12 months, the type of specialists visited, and the places of care were presented. It was found that children with a shunt (those with more severe type of SB), compared to those without a shunt, had more SB-related visits and specialist visits but not more ER or primary care visits. Although the generalizability of the findings to the greater population of children with SB is limited, results indicate that parents navigate a complex medical system to care for their child. Thus, it may be helpful to review systems of care for optimal structure to not only meet the health care needs of children with SB but also to support the families who care for them and, later, to meet and support the health care needs of adults with SB.

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Table 1

Characteristics of parent-respondents and their young children with spina bifida

	Total *n* = 101 (%)
Study Location	
Utah	73 (72)
Arizona	28 (28)
Parents' Characteristics	
Parental Status	
Biological parents	92 (2)
Adoptive parents	5 (5)
Legal guardian	1 (1)
Missing information	3 (3)
Sex	
Female	91 (90)
Male	7 (7)
Missing information	3 (3)
Children's Characteristics	
Sex	
Male	63 (62)
Female	38 (38)
Race/ethnicity	
Non-Hispanic White	68 (67)
Hispanic	28 (28)
Asian/Pacific Islander	4 (4)
Other	1 (1)
Spina Bifida Type	
Myelomeningocele	84 (83)
Lipomeningocele	12 (12)
Meningocele	5 (5)
Shunt Status	
Shunt	68 (67)
Without shunt	30 (30)
Missing information	3 (3)

Table 2

Place where child most often receives spina bifida or general/preventive medical care, as reported by parents

Place of Care	SB-Related *n* (%)	General or Preventive *n* (%)
Multidisciplinary clinic specialized in SB	64 (63.4)	9 (8.9)
Private physician's office	11 (10.9)	71 (70.3)
Clinic or health center (not multidisciplinary SB clinic)	7 (6.9)	13 (12.9)
Hospital outpatient department	6 (5.9)	1 (1.0)
Some other place	3 (3.0)	2 (2.0)
No one specific place most often	3 (3.0)	0 (0.0)
Emergency room	3 (3.0)	2 (2.0)
Do not know	1 (1.0)	0 (0.0)
Missing	3 (3.0)	3 (3.0)

Note. SB = spina bifida.

Table 3

Mean, standard deviation, and median number of children with spina bifida who visited a health care specialist in the previous 12 months by shunt status

	Shunt[1] *n* = 68				No shunt[1] *n* = 30				Total[1] *n* = 101			
Specialists Visits	***n***	**%**	**M (SD)**	**Md**	***n***	**%**	**M (SD)**	**Md**	***n***	**%**	**M (SD)**	**Md**
Urologist[2]	66	97.1	2.2 (1.8)	2	23	76.7	1.4 (0.8)	1	89	88.1	2.0 (1.6)	1
Neurosurgeon[2]	60	88.2	2.0 (1.6)	2	18	60.0	1.7 (1.7)	1	78	77.2	1.9 (1.6)	1
Orthopedist[2]	61	89.7	2.8 (2.2)	2	18	60.0	2.4 (1.9)	2	79	78.2	2.7 (2.1)	2
Neurologist	42	61.8	2.0 (1.7)	1	19	63.3	1.5 (1.6)	1	61	60.4	1.8 (1.7)	1
Ophthalmologist	28	41.2	1.4 (0.6)	1	11	36.7	1.4 (1.0)	1	39	38.6	1.4 (0.7)	1
Other												
Physical therapist	15	22.1	12.0 (13.0)	5	4	13.3	3.5 (4.4)	1.5	19	18.8	10.2 (12.2)	3
Occupational therapist	7	10.3	9.9 (13.0)	2	2	6.7	2.0 (0.0)	2	9	8.9	8.1 (11.8)	2
Ear nose throat specialist	6	8.8	2.5 (1.6)	2	2	6.7	0.5 (0.7)	0.5	8	7.9	2.0 (1.7)	1.5
Dietician/nutritionist	4	5.9	3.8 (2.6)	4	6	20.0	1.2 (0.4)	1	10	9.9	2.2 (2.0)	1
Cardiologist	5	7.4	1 (0.0)	1	–	–	–	–	5	5.0	1.0 (0.0)	1
Plastic or general surgeon	5	7.4	2 (1.7)	1	–	–	–	–	5	5.0	2.0 (1.7)	1
Gastrointestinal specialist	4	5.9	1.8 (0.5)	2	–	–	–	–	4	4.0	1.8 (0.5)	2
Psychologist	2	2.9	1 (0.0)	–	2	6.7	1.0 (0.0)	–	4	4.0	1.0 (0.0)	1
Orthodontist/pediadontist	2	2.9	1.5 (0.7)	–	–	–	–	–	2	2.0	1.5 (0.7)	–
Endocrinologist	1	1.5	2.0	–	–	–	–	–	1	1.0	2	–
Genetics specialist	1	1.5	1.0	–	–	–	–	–	1	1.0	1	–
Optometrist	1	1.5	1.0	–	–	–	–	–	1	1.0	1	–
Vision therapist	1	1.5	40.0	–	–	–	–	–	1	1.0	40.0	–
Missing	–	–	–	–	–	–	–	–	3	3.0	–	–

Note. Md = median, omitted when N < 3. M = mean. SD = standard deviation. Mean and median calculations include those who indicated ‘yes’ to a specialist visit in the previous 12 months. [1]Specialist visits and shunt data were missing for three participants. [2]Chi-square test of independence indicated an association between shunt status and visit to urologist, neurosurgeon, and orthopedist, $p < 0.01$.

Table 4

Number of parents educated by health providers about possible spina bifida-related complications

Complications	Shunt[1] (*n* = 68)	No shunt[1] (*n* = 30)	Total (*n* = 101)
Headaches	66 (97.1)	18 (60.0)	84 (83.2)
Vomiting	64 (94.1)	9 (30.0)	73 (72.3)
Fever	60 (88.2)	12 (40.0)	72 (71.3)
Difficulties eating	47 (69.1)	11 (36.7)	58 (57.4)
Fussiness	47 (69.1)	11 (36.7)	58 (57.4)
Difficulties breathing	35 (51.5)	9 (30.0)	44 (43.6)
Gagging	33 (48.5)	6 (20.0)	39 (38.6)
Weak cry	19 (27.9)	6 (20.0)	25 (24.8)
Arm weakness	21 (30.9)	7 (23.3)	28 (27.7)
High pitched cry	16 (23.5)	4 (13.3)	20 (19.8)
Noisy breathing	22 (32.4)	9 (30.0)	31 (30.7)
Cyanosis (turning blue)	23 (33.8)	8 (26.7)	31 (30.7)
Other	38 (55.9)	14 (46.7)	52 (51.5)
Don’t know	0 (0.0)	1 (3.3)	1 (1.0)

Note. Question on the survey given to parents was: *Infants and toddlers with spina bifida can experience additional complications that need medical care. I am going to read you a list of items, please let me know whether or not any of your health care providers have told you to watch out for any of these symptoms in “X.”* [1]Shunt status was missing for three children.