Pediatric short-bowel syndrome: the cost of comprehensive care

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ABSTRACT
Background: Little information is available about the financial charges incurred by patients with short-bowel syndrome (SBS). This is particularly true for pediatric SBS patients who receive some of the most complex medical care.

Objectives: The aims of this study were to determine the total cost of care for these patients and to analyze their utilization of home and hospital-based health care services.

Design: This was a retrospective review of the total charges incurred by 41 children with SBS over the past decade, encompassing both inpatient and home-care charges.

Results: The mean (± SD) total cost of care for pediatric SBS was US$505 250 ± US$248 398 (corrected for inflation to the year 2005) for the first year of care alone. Inpatient hospitalization accounted for most of these expenses (US$416 818 ± US$242 689, or 82% of the total), and this was attributable to prolonged requirements for intensive care resources, numerous surgical procedures, and multiple readmissions during the first year of diagnosis. Hospital-based costs steadily declined in subsequent years, but home-care services, in stark contrast, unexpectedly increased every year for the first 5 y of diagnosis—a trend that was highly significant (P < 0.005), reaching US$184 520 ± US$111 075 for the fifth year of home care. This increasing cost was attributable to increasing complications of parenteral nutrition, especially infectious complications. Although per-patient charges varied widely, the mean total cost of care per child over a 5-y period was US$1 619 851 ± US$1 028 985. A strong correlation was found between higher charges and infants with <10% of predicted small-bowel length.

Conclusions: This study was the first to calculate the total costs for pediatric SBS patients and to provide an in-depth analysis of these patients’ actual utilization of health care services. This information may help guide health care providers and families who have children with SBS. The comprehensive care of pediatric SBS patients costs significantly more than what has been previously estimated. Contrary to previous views, home care significantly increases each year after diagnosis. Am J Clin Nutr 2008;88:1552–9.

INTRODUCTION

Short-bowel syndrome (SBS) is a devastating disease process in infants and children with mortality rates persistently reported in the 20–30% range despite marked improvements in the care of these children (1–4). Care for SBS patients is complex and resource-intensive, often beginning at the time of birth or shortly thereafter, and requires prolonged admission to an intensive care unit, multiple surgical procedures (5) within the first year of life, and specialized nutritional support (6–10). Treatment of such patients may be required for several years and generally involves a multidisciplinary approach, including inpatient care, home infusion services, home care, nursing visits, and clinical appointments (10–15). Approximately 40 000 patients receive home parenteral nutrition (PN) in the United States (16). Although children account for a small number of SBS patients, the monetary cost associated with the care of this group of patients can be sizeable.

No comprehensive study to date has examined the actual costs of caring for pediatric SBS patients over a defined period of time. The cost of caring for pediatric patients receiving home PN has been estimated from adult data (17); however, these adult patients were not exclusively SBS patients. Furthermore, the low number of hospitalizations in that particular report suggests that the adult patients required far less care than many pediatric patients require. In fact, the actual total cost of care (COC) for pediatric SBS remains unknown (18, 19). Although estimates have been made, little to no substantive information is actually available (20), and many of these monetary estimates date back to almost 3 decades ago (21, 22), which makes the extrapolation to current clinical practice difficult. Additionally, the etiologies and comorbidities of pediatric SBS differ significantly from those for adult SBS patients (23–25). These patients often require not only intensive hospital treatment, but also a sustained program of home-based care for several years, especially PN, until intestinal function is restored. Knowing the total COC may significantly affect the welfare of patients and their families and potentially influence decisions regarding therapeutic options; this is particularly important because many third party payors have strict capitations on life expenditures for medical insurance. Finally, it is important for government services to better understand the economics of SBS. Current limitations on reimbursement for such patients may grossly underestimate the actual financial costs required to provide adequate care for such children (26).

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The lack of comprehensive data on the total COC stems from the fact that pediatric SBS requires health care services in both the home and in the hospital setting over a protracted period of time. Few investigators have had access to this type of economic data. Our health system has the very unique ability to track a large number of pediatric patients over the past decade who received the vast majority of their care from our University of Michigan Health Service. This allowed us to calculate a comprehensive estimate of the total charges incurred for these patients, both during their inpatient hospitalizations and over the duration of all home care. This study gives the most comprehensive picture of these charges to date and should provide very useful information for health care providers who care for pediatric SBS patients.

SUBJECTS AND METHODS

Patient population

Pediatric SBS is defined as the loss of ≥70% of small intestinal length from surgical resection or congenital defect or ≥2 mo of PN dependence due to complete intestinal dysfunction. Standard, commercially available PN formulas were used. The onset of the diagnosis of SBS was always considered the reference point (time = zero). Between March 1992 and January 2005, 117 pediatric SBS patients were cared for at the CS Mott Children’s Hospital of the University of Michigan. Of this group, 41 families elected to receive their postdischarge home care through HomeMed at the University of Michigan Health System—a home infusion pharmacy service operated by our hospital system. These patients also utilized our University home health agency and had their follow-up clinic visits at our Intestinal Failure Clinic. For any 1 y of care, the total charges for these 41 patients (referred to as COC in this report) for each of these patients could be calculated for their care.

This study was a retrospective review of all in-hospital and home-care charges during the study period and was conducted in accordance with the ethical standards of the University of Michigan Institutional Review Board (IRB) and was approved by this Board (approval 2000-0254).

Calculation of charges

All charges were converted to 2005 US dollars using inflation correction factors published by the US Department of Labor’s Bureau of Labor Statistics. Charges were rounded to the nearest dollar after the calculations were made. In-hospital charges were defined as all inpatient hospitalizations (including procedural charges, diagnostic testing, and hospital room charges), as classified by DRG code and procedural code. The COC for this work represents the allowable billable charges as set by Medicaid. Additionally, because of the nature of our billing system, all out-patient clinic visits and outpatient diagnostic test charges that were drawn in our clinic were included in the in-hospital portion of the charges. Home-care charges were defined as all care provided in the home, including both home infusion charges and nursing visits, as classified according to an internal system.

Home infusion charges included all therapies performed in the home with the exclusion of nursing visits. Briefly, home-care charges were placed into 1 of 5 categories: antimicrobial medications, other parenteral medications, intravenous fluid therapy (nonnutrition), enteral nutrition therapy and accompanying products, and PN. Costs associated with supplies, pumps, tubing, and accessories were placed into the corresponding category. Prescribed medications filled by the patient’s family pharmacy were not included in these home infusion figures. In general, nursing visit charges were low and accounted for ≤2% of net charges. Most nursing visit charges were incurred within the first year of care. Because of this, nursing visit charges were incorporated in the total COC under the subheading of home care. The duration of each type of therapy in days, as well as in the year after the diagnosis of SBS, was recorded for each charge. All analyses were conducted from the purchaser’s perspective; that is, the charges calculated were the price that a third-party payor would pay for the services rendered. Charges in each case followed those prescribed by Medicare. Nonreimbursed charges were not included in the analysis, so that results describe only the actual charges associated with pediatric SBS.

We looked at the first 5 y of SBS, calculating the total COC per year for the first 5 y. We chose this time frame because most patients who survive SBS achieve independence from PN within the first 5 y, although some continue to remain PN-dependent.

Definitions

SBS was defined as a loss of ≥70% of small intestinal length from surgical resection or congenital defect or ≥2 mo of PN dependence because of complete intestinal dysfunction. The onset of the diagnosis of SBS was always considered the reference point (time = zero). Thus, patients treated at different times throughout the study period could be directly compared at equivalent points in time subsequent to the development of SBS (onset). In addition, not all patients elected to continue their home care through the University of Michigan for the entire duration of the study. Therefore, data were included only for those years when a patient was fully serviced by our institution. Finally, once a patient was completely weaned off of PN and had their intravenous line removed, we considered the patient to be terminated from the study. Because this group of patients was not a constant number, the various shifts in patient numbers throughout the study are conveniently provided in Table 1.

Statistical methods

Charges for all patients during the first through the fifth year after onset of SBS were aggregated for each year, and means were compared with analysis of variance (ANOVA). The least-squares method was used for post hoc analysis to detect statistical

<table>
<thead>
<tr>
<th>TABLE 1</th>
<th>Breakdown of the outcomes of all 41 patients reported in this study1</th>
</tr>
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<tbody>
<tr>
<td>Year after onset of SBS</td>
<td>No. of patients at start of year on PN</td>
</tr>
<tr>
<td>1</td>
<td>41</td>
</tr>
<tr>
<td>2</td>
<td>23</td>
</tr>
<tr>
<td>3</td>
<td>17</td>
</tr>
<tr>
<td>4</td>
<td>122</td>
</tr>
<tr>
<td>5</td>
<td>8</td>
</tr>
</tbody>
</table>

1 Note that the results for each year represent the status of patients at the end of each year. SBS, short-bowel syndrome; PN, parenteral nutrition.

2 Two patients were <4 y of age and therefore were not included in the data for the following year.

3 Follow-up of these 7 patients showed that 4 were successfully weaned from PN and 3 died.
differences between groups. \( P < 0.05 \) was considered significant. Data are expressed as means ± SDs; medians are provided in some cases.

Covariates that could potentially influence the amount of total charges were examined by using linear multivariate regression analysis. Because many patients did not survive past the first year, and the greatest costs were incurred during this time period, this multivariate analysis was confined to the first year after the diagnosis of SBS. In the initial linear regression model, 3 covariates were considered (survival, cholestasis, and the estimated percentage of the remaining small intestine). Because the percentage of the remaining small-bowel length varied depending on the gestational age of the child when initially measured, we used a conversion method to estimate the percentage of predicted small bowel remaining based on gestational age, as opposed to a set number of centimeters (4, 27). Covariates that were found not to be significant were subsequently removed by backward elimination (28, 29). Statistical analysis was performed with SPSS version 13.0 (SPSS Inc, Chicago, IL).

RESULTS

Demographics

Of the 41 pediatric SBS patients, 16 (39%) were boys. The onset of SBS ranged from 0 d to 17.7 y. Onset of SBS occurred at the time of birth in 21 infants (congenital defects, including gastroschisis and severe intestinal atresia), within 2 mo of birth in 12 infants (predominantly due to necrotizing enterocolitis), and between 5 mo and 17.7 y of age in the remaining 8 patients. SBS was due to volvulus, trauma, and miscellaneous etiologies in these older patients. None of the patients had a known malignancy. The study population was representative of the spectrum of etiologies of pediatric SBS (Table 2). With the onset of SBS, all 41 patients required total PN support. In comparison with a larger group of 117 infants from a previous study (4), no significant differences were noted between the distribution of the diagnoses, gestational age (35.6 ± 0.9 wk in this series compared with 33 ± 4.7 wk in the larger group), and overall survival (30% for the current study and 27.5% for the larger group of SBS patients).

Duration of follow-up

All data were stratified according to the year after onset of SBS. Data were analyzed for each patient for the years when the patient received both home care and hospital care at our institution. The number of patients available for analysis during each year declined with patient deaths and attrition due to weaning off of PN (Table 1). Some patients were referred to our institution after having received care elsewhere, and their data were analyzed only from the years when all their medical care was provided at our institution. As stated above, once patients were receiving 100% enteral supplementation, they were considered to have completed contributing data for this study. Although home care may still have been provided, the level of intensity declined dramatically, and many of these patients continued with long-term enteral supplementation. Finally, because of the sizeable attrition of patients over the first 5 y after the onset of SBS, only data for the first 5 y after the onset of SBS were included in the analysis, even though some of the patients received PN for longer periods of time.

TABLE 2

<table>
<thead>
<tr>
<th>Diagnosis</th>
<th>No. of patients</th>
<th>Percentage of total (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gastrostschis</td>
<td>11</td>
<td>26.8</td>
</tr>
<tr>
<td>Intestinal atresia</td>
<td>6</td>
<td>14.6</td>
</tr>
<tr>
<td>Volvulus</td>
<td>7</td>
<td>17.1</td>
</tr>
<tr>
<td>Necrotizing enterocolitis</td>
<td>9</td>
<td>22.0</td>
</tr>
<tr>
<td>Miscellaneous</td>
<td>8</td>
<td>19.5</td>
</tr>
</tbody>
</table>

Frequency and duration of admission to hospital

During the first year after the onset of SBS, frequent readmissions were observed for most patients. On average, patients required 5.8 ± 3.4 readmissions after the initial hospitalization during the first 12 mo alone (median: 6 readmissions; range: 1–12 readmissions). This resulted in a mean (± SD) of 119 ± 61 in-hospital days in the first year after diagnosis of SBS (per patient). The duration of the initial hospitalization (at onset of SBS) averaged 67.1 ± 50.3 d (median: 44 d; range: 19–161 d). However, the duration of hospitalizations for subsequent readmissions during the first year were much shorter (10.0 ± 14.7 d; median: 5 d) per admission (\( P < 0.0001 \)). The frequency of readmission, although initially very high, also steadily decreased with time (Figure 1). This trend in the decline in readmission rates was highly significant (\( P < 0.0001, R^2 = 0.762 \)).

Total cost of care

A summary of the total COC for the 5 y of the study is shown in Figure 2. The COC is summarized as mean dollar values. A wide SD is noted, which reflects the diversity of this patient population. COC was greatest in the first year, amounting to >US$500 000. Also interesting was that the COC for subsequent years was fairly stable, between ~$250 000 and $300 000/y (median: $296 808/y). We next broke down these charges between in-hospital and home care.
In-hospital admission charges

Overall, for the entire 5-y period of study, the average pediatric SBS patient spent a mean (± SD) total of 146 ± 188 d in the hospital (median: 82 d; range: 14–1064 d) and incurred a total cost of $1 619 851 ± $1 028 985, inclusive of both hospital and home-care costs. However, hospital costs varied widely between patients, and therefore a more detailed analysis was performed. Data from each patient were stratified according to the year post-onset of SBS.

The total in-hospital COC values per year are shown in Table 3. Not surprisingly, these costs diminished each year, as the total number of hospitalizations and the duration of hospitalizations declined annually (Figure 1); mean in-hospital charges declined to <$50 000 by year 5. Apart from minor fluctuations, there was no significant change in the daily COC of inpatient hospitalizations over the course of time.

Home-care charges

A summary of all home-care patient charges is given in Table 3, including the contrast between inpatient charges and home-care charges during these same years. What is particularly striking about the data is the fact that home-care costs continued to rise each year. Although the inpatient COC trended steadily downward during these 5 y, as mentioned above, the total home-care COC continued to increase every year after the onset of SBS. Home-care COC started at $87 932 ± 70 079 (median charge: $81 160) for the first year and rose to $184 520 ± 111 075 by year 5 (median charge: $202 980). Note that data reflect an annual, not a cumulative, average. Thus, for patients who continue to utilize home-care services for SBS care (including enteral supplements), annual costs continued to rise. A more detailed breakdown of home-care charges is given in Table 4. This breakdown shows the causes of this persistent increase in home-care charges. Specifically, the persistently high COC for PN, along with increasingly high charges for other therapies (particularly antimicrobials), helped to explain this rise in home-care charges. This rise in charges occurred despite the fact that many infants were actually being weaned off of PN during this time period.

Outcome of patients and relation of survival and length of the small bowel to COC

The duration of PN dependence ranged from 6 mo to 11.6 y, and all patients who were eventually weaned off PN (defined as maintaining adequate growth on enteral nutrition alone) survived throughout the duration of the study. In contrast, all patients who died remained dependent on PN up to the time of death. Of the patients who died, the mean survival was 4.3 ± 5.7 y (range: 6 mo to 11.6 y).
to 19 y; median: 1.3 y). Two patients who remain PN-dependent were still alive at the last follow-up at age <5 y. The time required to wean off of PN (enteral independence) in the group that ultimately did was 2.6 ± 3.1 y (range: 2.4 mo to 12.6 y of PN dependence; median: 1.5 y). The data provided in Table 1 better clarifies the patients analyzed in this study.

On the basis of our finding of an almost 30% mortality rate, we next stratified our patients as survivors and nonsurvivors. The annual COC for patients in the group who ultimately died, compared with those who survived, is shown in Figure 3. The striking difference between patients who ultimately survived and those who did not is that the annual COC was always greater in the nonsurviving group. The mean annual cost of in-hospital care for patients in the group that survived, compared with the group that did not survive, are shown in Figure 3B. Aside from the first year, hospital costs were consistently greater in the group that ultimately died, and the difference became more pronounced in subsequent years. This was thought to be due to the fact that the survivors had a dramatic decrease in their annual hospital costs with time. As can be seen, hospitalization cost data after the second year appeared to be a strong independent predictor of the ultimate outcome. Home-care costs are shown in Figure 3C. Remarkably, even though patients who died spent a great deal of time in the hospital, their home-care costs were also consistently higher than those for the patients who ultimately survived. The data shown included only those children who continued to utilize home care.

Similar to the observations of others (30), our group has previously shown that patient survival could be predicted by bowel length. Our own previous work also showed a correlation between those with <10% of their predicted small-bowel length and an inability to wean off PN (4). The total COC per bowel length is shown in Figure 4. Those patients with <10% of predicted small-bowel length had a consistently greater COC for all of the years studied. These higher costs were predominately due to the fact that these patients had a greater frequency of inpatient hospitalization and a greater utilization of hospital resources. As was previously shown, patients with <10% of the estimated normal small-bowel length remaining were significantly more likely to remain permanently PN-dependent and are at a much greater risk of morbidity and mortality. For the entire 5-y analysis, these patients incurred an almost doubling of charges: $2 029 958 ± 1 044 620 compared with $1 301 026 ± 862 123

Regression analysis of charges during the first year of SBS

To better define covariates that may predict a greater COC, a regression analysis was done for patients in the first year after SBS. Interestingly, nonsurvival (nonstandardized β coefficient: 73 807; standardized coefficient: 0.113; P = 0.719) and the development of cholestasis (defined as conjugated bilirubin >2.5 mg/dL; nonstandardized β coefficient: −84 756; standardized coefficient: −0.124; P = 0.606) were not found to be significant predictors of greater COC. This was interesting because these 2 factors are often thought to incur the greatest complexity of patient care. However, a strongly positive correlation was found between those children with small-bowel lengths <10% of that predicted for gestational age and higher patient charges (P < 0.003). This significance also persisted despite the removal of nonsurvivors (nonstandardized β coefficient: 507 403 ± 225 194; standardized coefficient: 0.623; 95% CIs: 28 177, 1 047 208; P = 0.003). Univariate analysis of other factors that could potentially influence survival, including the specific etiology of SBS, the presence or absence of an ileocecal valve, and the total number of septic episodes, showed no significant correlation.

DISCUSSION

The care of patients with SBS is complex, is expensive, and requires a long-term commitment by trained individuals (3, 31–33). Predicting which patients will survive and their overall outcomes was addressed well over 3 decades ago (34). Recently, several evaluations of large pediatric intestinal failure centers have identified survival rates of ≈70–80% (4, 13, 35). Risk factors for survival in these series include the overall length of retained small bowel, the presence or absence of an ileocecal valve, and whether or not the child develops PN-associated liver disease. Unfortunately, many of these patients may require long-term PN for years. The care of a child with SBS is particularly challenging, and, unlike in adults, is associated with a higher rate of liver injury and difficulty in maintaining vascular access (10, 11, 13–15, 31, 36–39).

Our study addressed a critical issue regarding the care of these children—the overall financial charges that pediatric SBS patients incur during their care. Although previous reports have estimated the COC to be between $125 000 and $250 000/y (18, 19, 40), most of these are estimates are based primarily on data for adult patients. Furthermore, most of these values are not based on complete data sets, but rather on estimations based on idealized patients. Our findings show that the cost over the first 5 y after the onset of SBS is greater than some of these previously estimated values—exceeding $1.6 million for the first 5 y of treatment and ranges from 1.3 to 2.0 million dollars if PN is required for all 5 y. The study also showed that charges were predominately inpatient during the first year, and these charges

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**TABLE 4**

Breakdown of the most common home-care charges by therapy type for 2005

<table>
<thead>
<tr>
<th>Therapy</th>
<th>Year 1</th>
<th>Year 2</th>
<th>Year 3</th>
<th>Year 4</th>
<th>Year 5</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Antimicrobials</strong></td>
<td>11 887 ± 11 766</td>
<td>19 141 ± 19 161</td>
<td>23 996 ± 32 431</td>
<td>27 792 ± 25 371</td>
<td>34 784 ± 33 746</td>
</tr>
<tr>
<td><strong>Injectables</strong></td>
<td>4242 ± 5359</td>
<td>5983 ± 6622</td>
<td>17 716 ± 23 992</td>
<td>9631 ± 7020</td>
<td>16 951²</td>
</tr>
<tr>
<td><strong>Intravenous hydration</strong></td>
<td>6149 ± 5827</td>
<td>7187 ± 6053</td>
<td>13 473 ± 26 876</td>
<td>16 205 ± 26 031</td>
<td>7777 ± 5344</td>
</tr>
<tr>
<td><strong>Enteral nutrition</strong></td>
<td>5827 ± 5538</td>
<td>11 248 ± 9384</td>
<td>13 917 ± 10 742</td>
<td>16 319 ± 11 706</td>
<td>21 288 ± 10 637</td>
</tr>
<tr>
<td><strong>Parenteral nutrition</strong></td>
<td>83 262 ± 59 085</td>
<td>106 154 ± 64 118</td>
<td>152 411 ± 51 522</td>
<td>147 035 ± 61 274</td>
<td>126 579 ± 82 124</td>
</tr>
</tbody>
</table>

¹ All values are ± SD. Costs are in US dollars corrected for inflation to the year 2005.
² No SD is given because the amount reflects a small number of patients.
accounted for a large portion (≈60%) of all charges incurred. However, we also unexpectedly found that the cost of home care for these children continued to rise over time. This rise in home charges occurred despite predicted improvement for many of these children during the same time period. This suggests that, although there are numerous benefits to patients receiving home care, costs for such care remain a substantial financial burden to the patients’ families and insurance providers.

The cost of delivery of PN in the United States is a difficult number to determine because of the large number of medical providers and insurance payors. Additionally, costs of PN in the United States appear to be disproportionately higher (4-fold) than those in Canada and the United Kingdom, which makes an extrapolation of costs from other countries difficult (41). An estimate based on Medicare expenditures from 1989 to 1992 showed that yearly costs rose to 137 million dollars by the end of the analysis period (16). Extrapolation of comprehensive costs of pediatric SBS from these numbers is not possible because these costs were only for home PN, were primarily based on adult data, and did not include the hospital-associated costs of pediatric SBS. Furthermore, children with SBS (based on the diagnosis “congenital disorders”) made up <2.5% of the total population analyzed. A recent comprehensive study of adult home PN patients indicated costs that were markedly less than those found in our current study. In that study, Reddy and Malone (17) found that costs (also based on charges) varied considerably between patients. The mean PN charges were only $70 000 y, and mean annual charges for enteral nutrition were merely $18 000. Importantly, that study only dealt with patients older than 18 y, again emphasizing the fact that pediatric SBS care and costs may substantially differ from those of adult SBS. Additionally, the indications for receiving home PN and enteral nutrition in that study were not necessarily for SBS; thus, the charges probably do not fully reflect the complex medical conditions associated with pediatric SBS patients. For example, the average number of hospitalizations in Reddy and Malone’s study was between 1 and 2 y—far fewer than in our series of pediatric patients. Another
limitation of this study was that charges were estimates based on 1996 Medicare allowable charges, whereas the results in our study represented actual charges submitted to third-party payors. Although these costs were probably similar (aside from the 9-y difference in inflationary correction), we believe our study allowed for a more accurate assessment of patient charges for pediatric SBS.

In-hospital charges accounted for more than one-half of all charges. In-hospital charges directly correlated with the duration of hospital days. Thus, in-hospital charges were greatest in the first year after SBS and progressively decreased with time. Clearly, the charges incurred by these patients accounted for the bulk of their initial hospitalization, which was usually in an intensive care unit setting and was quite lengthy. It is well-established that home PN administration is economically much less expensive than in-hospital PN (42, 43). Therefore, it was interesting that home-care charges continued to rise through year 5 of the study. A major component of this rise was the fact that care shifted from the predominant inpatient side to home care over the course of care. Nevertheless, if this was the only cause, then it would seem that charges would plateau after year 3. We suspect that despite the charges being corrected for inflationary factors, the cost of medical care continued to rise in a greater proportion throughout the study, and this would be reflected in this progressive rise in charges over time. The causes of these increased charges are not completely understood. It may also be that as patients grow and age they may transition to a higher requirement of PN (ie, 1- to 2-L volumes of infusion). Additionally, more complex antimicrobials (ie, liposomal-based antifungal therapy) may also contribute to these higher charges. The fact that the combined charges of in-hospital and home care equaled or exceeded $250 000/y, even after the fifth year after onset of SBS, emphasizes the fact that previous estimates of the COC for SBS patients have underestimated the ongoing costs of care for pediatric SBS when compared with the charges actually incurred.

One potential, albeit small, source of error may arise from the fact that we included outpatient clinic charges and the costs of blood draws in the total hospital charges instead of in the home-care charges. This was done simply because these charges are billed through the university hospital accounts. Because of the nature of our billing system, all outpatient clinic visits and out-patient diagnostic test charges that were drawn in our clinic were included in the in-hospital portion of the charges. Although doing this may detract from total home-care charges, in fact, these visits accounted for <1% of all yearly COC. For example, 6 yearly clinic encounters (level 3 or 4 charges of care × ≈$150) would amount to $900/y. Therefore, we simply kept the original hospital data intact, because this small factor would not significantly change the overall result of the study. Likewise, no significant impact was made on home-care charges.

Costs in this study were estimated for a cohort of 41 infants and children. Although this was a small study, we believe that our patient population accurately reflected a broad spectrum of SBS patients. The overall survival in our current report mirrored a recent large series of >100 SBS infants and children also reported by our Intestinal Failure Center and other recent series (2, 4, 7, 10). Additionally, similar to other reports of children with SBS, the ability to gain enteral independence was noted in more than two-thirds of our patients (4, 10, 30). Of note was the fact that many of our children gained such independence after several years of home PN. This is noteworthy because several other authors have expressed doubt that any patient with SBS is likely to be weaned off of PN after 3 y of PN dependence (44, 45). Importantly, these reports are in adults. The data strongly suggest that children have a much greater capacity for intestinal adaptation and regeneration after SBS than do adults. In our cohort of patients, we observed 8 patients (out of 41) with a diagnosis of SBS who were PN-dependent for a minimum of 3 y, who ultimately were successfully weaned from PN >3 y after the onset of SBS; 3 were weaned off PN even after having received it for 5 y. Intensive nutritional care of infants and children with SBS has been shown to be effective in weaning such patients from PN who otherwise would have been maintained on this therapy (9).

It was interesting to note that those patients with a small-bowel length <10% of predicted for their gestational age incurred the highest charges. The mean charges exceeded 2 million US dollars—a level that typically exceeds the lifetime maximum benefits of many third-party payors. Thus, families, and medical providers may incur a large portion of this financial responsibility. This high financial cost may also place a significant burden on many hospitals because ≈15% of patients younger than 18 y will lack insurance coverage for at least a portion of the time (46). Thus, our findings may require a rethinking about the current provider capitations on medical and surgical care. In a recent analysis of survival in pediatric SBS patients, we found that those patients with a small-bowel length <10% of predicted had a significantly lower survival than did those infants with longer bowel lengths (4) (47). Conceivably, this high-risk group of patients may possibly need to be identified early on for alternative therapies such as intestinal transplant assessment, because determination of which patients may be candidates for transplantation has been a challenge (48). It was also noteworthy that other covariates in our study did not predict higher patient charges, including the development of cholestasis and whether or not the patient eventually died of SBS. It is, however, possible that the early death of some of these patients resulted in a low total amount of charges, which offset higher charges from these generally more complex patients. It is conceivable that as the care of SBS patients becomes further coordinated (14, 15), as surgical techniques become further advanced (49), as additional medical therapies are used (50, 51), and as mortality rates continue to decline (13), the overall COC for such patients may well decline in future assessments of such patients.

In conclusion, this study was one of the first to comprehensively examine the true cost of caring for a child with SBS. The yearly costs of care are high, and home care continues to increase with each year the patient remains on PN, despite the fact that this care is predominately in a home setting after the first year after the onset of SBS. The data also provide further support for the financial benefit of providing this care at home rather than on an inpatient basis. These numbers may prove useful when attempting to evaluate the financial burden a family faces with a child with such a diagnosis. The data may also suggest a rethinking of financial capitations that may be placed on such patients.

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