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SACRAL NERVE STIMULATION FOR CONSTIPATION AND FECAL INCONTINENCE IN CHILDREN: LONG-TERM OUTCOMES, PATIENT BENEFIT, AND PARENT SATISFACTION

Peter L. Lu, Ilan J.N. Koppen, Danielle K. Orsagh-Yentis, Karen Leonhart, Erica J. Ambeba, Katherine J. Deans, Peter C. Minneci, Steven Teich, Karen A. Diefenbach, Seth A. Alpert, Marc A. Benninga, Desale Yacob, Carlo Di Lorenzo

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ABSTRACT

Objective: To evaluate the long-term efficacy of sacral nerve stimulation (SNS) in children with constipation and describe patient benefit and parent satisfaction.

Methods: Using a prospective patient registry, we identified patients <21 years old with constipation treated with SNS for >2 years. We compared symptoms, medical treatment, PedsQL Gastrointestinal Symptom Scale (GSS), Fecal Incontinence Quality of Life Scale (FIQL), and Fecal Incontinence Severity Index (FISI) before SNS and at follow-up. We contacted parents to administer the Glasgow Children's Benefit Inventory (GCBI) and a parent satisfaction questionnaire.

Key Results: We included 25 children (52% male, median age 10 years): 16 had functional constipation, six anorectal malformation, two tethered spinal cord, and one Hirschsprung’s disease. Defecation frequency did not change after SNS but patients reporting fecal incontinence decreased from 72% to 20% ($P<.01$) and urinary incontinence decreased from 56% to 28% ($P=.04$). Patients using laxatives decreased from 64% to 44% (ns) and patients using antegrade enemas decreased from 48% to 20% ($P=.03$). GSS, most FIQL domains, and FISI were improved at follow-up. Six (24%) patients had complications requiring further surgery. Of the 16 parents contacted, 15 (94%) parents indicated positive health-related benefit and all would recommend SNS to other families.

Conclusions & Inferences: Sacral nerve stimulation is a promising and durable treatment for children with refractory constipation, and appears particularly effective in decreasing fecal incontinence. Although a quarter of patients experienced complications requiring additional surgery, nearly all parents reported health-related benefit. Future studies to identify predictors of treatment response and complications are needed.
INTRODUCTION

Constipation is a common problem in children, with an estimated worldwide prevalence of 12%. While the majority of children respond to medical and behavioral treatment, a sizable portion of these children continue to have symptoms despite conventional treatment. Unfortunately, treatment options for children with constipation refractory to conventional treatment are limited. The most recent guidelines from the European and North American pediatric gastroenterology societies on the evaluation and treatment of functional constipation in children recommend consideration of anal sphincter botulinum toxin injection, transanal irrigation, antegrade continence enemas (ACE), sacral nerve stimulation (SNS), and partial or total colonic resection for treatment of intractable constipation.

Sacral nerve stimulation involves low-amplitude electrical stimulation of the sacral nerve via an electrode placed through the sacral foramen. Over the past two decades, experience with the use of SNS to treat adults with constipation and fecal incontinence has grown, and SNS is now considered the first-line surgical treatment for adults with fecal incontinence refractory to conventional treatment. However, recent studies have not shown SNS to be an effective treatment for adults with constipation. Experience with the use of SNS in children with constipation has been positive thus far but remains limited, and the long-term outcomes of SNS treatment in children with constipation are not yet clearly understood. Although SNS appears to be a promising treatment option for children with refractory constipation, an understanding of its long-term efficacy and safety in children is needed before this treatment modality becomes more widely accepted. Therefore, the objective of our study was to describe the long-term outcomes of children with constipation treated with SNS by evaluating changes in symptom severity and quality of life, perceived health-related patient benefit, and parent satisfaction.

MATERIALS AND METHODS

We performed a prospective observational cohort study. We included patients up to 21 years of age with constipation who underwent SNS initiation at Nationwide Children’s Hospital in Columbus, OH, USA between May 2012 and November 2013 and therefore had been treated with SNS for at least 2 years at the time of data collection in November 2015. We recorded information on patient symptoms, laxative and ACE usage, patient-reported outcomes, relevant diagnostic test results, and complications of SNS at baseline and at each follow-up visit after SNS initiation. Patient-reported measures of symptom severity and quality of life included the PedsQL Gastrointestinal Symptom Scale (GSS), Fecal Incontinence Quality of Life Scale (FIQL), and Fecal Incontinence Severity Index (FISI).
This information was entered into a patient registry using the REDCap© electronic data capture tool. Successful response to SNS was defined as having >2 bowel movements and <1 episode of fecal incontinence per week at follow-up.

Three members of our research team who were not directly involved in the clinical care of SNS patients contacted each patient’s family by telephone in November 2015. We asked to speak to a parent or guardian of each patient. After a brief description of our study, we asked for verbal consent for participation in our study. If there was no answer, we left a voicemail message requesting a return call. We contacted each family up to three times. For those who agreed to participate, we administered the Glasgow Children’s Benefit Inventory (GCBI), a measure of health-related patient benefit after an intervention. The GCBI score ranges from −100 to +100, and positive scores indicate perceived benefit. We also asked two questions relating to satisfaction with the SNS treatment: (1) If you were able to go back in time to before your child started SNS treatment, would you still decide to proceed with SNS treatment? (2) Would you recommend SNS treatment to another family with a child who has the same symptoms as your child? Families were asked to explain their reason for each answer and responses were recorded by members of the research team. The Institutional Review Board approved our study protocol.

Sacral nerve stimulation procedure

Sacral nerve stimulation treatment was initiated in two stages for all patients. Both stages were performed under general anesthesia with the patient in the prone position. The first stage procedure involved placement of a tined lead at the S3 sacral nerve root under fluoroscopic guidance. Placement was confirmed by observation of a bellows response of the pelvic floor and plantar flexion of the great toe with stimulation. The lead was then connected to a temporary pulse generator that remained external to the patient. Symptoms were monitored for a 2-week period. If clinical improvement in constipation was observed, the patient then underwent the second stage procedure, which involved implantation of a permanent pulse generator into the subcutaneous tissue of the buttock. The InterStim® System (Medtronic, Inc., Minneapolis, MN, USA) was used for all patients. Sacral nerve stimulation procedures were performed either by a pediatric surgeon (ST) or a pediatric urologist (SAA). Patients generally returned for an initial follow-up appointment 2-4 weeks after the second stage procedure, followed by appointments at 3-month intervals through the first year after SNS initiation. Subsequent follow-up appointments generally occurred at 3-6 month intervals.
Data collection and analysis
For all patients meeting our inclusion criteria, we reviewed encounters at baseline prior to SNS initiation and at their most recent follow-up. Data are presented as medians and interquartile ranges for continuous data and as frequencies and percentages for categorical data unless otherwise specified. We compared patient symptoms, laxative and ACE usage, GSS, FIQL, and FISI at baseline and follow-up. McNemar’s test and Wilcoxon signed-rank test were used for comparison as appropriate. We also compared outcomes between patients with and without concurrent fecal incontinence using Pearson’s chi-squared test and Wilcoxon rank-sum test. P-values less than .05 were considered statistically significant. Statistical analyses were performed using SAS 9.3 (SAS Institute, Inc., Cary, NC, USA) and SPSS (SPSS Statistics for Windows, Version 22.0, Armonk, NY, USA).

RESULTS
We included 25 patients in our study (52% male, median age at SNS initiation 10 years, range 6-19 years). All of the patients who completed the first stage procedure during the time period studied (between May 2012 and November 2013) went on to complete the second stage permanent implantation. At the time of data collection, patients had been treated with SNS for a median of 2.3 years (IQR 2.1-2.7). Sixteen patients (64%) had functional constipation according to the Rome III criteria, six (24%) had a history of anorectal malformation, two (8%) had tethered spinal cord, and one (4%) had Hirschsprung’s disease. The indication for SNS treatment in all patients was severe constipation. Both patients with tethered spinal cord had undergone surgical detethering prior to SNS initiation, one undergoing detethering five years prior and the other two years prior. Both continued to have constipation despite either ACE treatment or an aggressive laxative regimen. The patient with Hirschsprung’s disease had incomplete defecation and retentive fecal incontinence despite laxative treatment. At baseline, 18 patients (72%) had concurrent fecal incontinence, 14 (56%) had urinary incontinence, and two (8%) had urinary retention.

Clinical symptoms
As shown in Table 1, the majority of patients had three or more bowel movements per week at baseline, and this did not change significantly with SNS treatment at follow-up. However, the presence of both fecal incontinence and urinary incontinence decreased significantly after SNS treatment ($P < .01, P = .04$). Seventeen of 25 patients (68%) had >2 bowel movements and <1 episode of fecal incontinence per week at follow-up and therefore fulfilled our criteria for successful response. Ten of these 17 patients had functional constipation and the remaining seven patients had organic causes of constipation.
Chapter 15

Constipation treatment

The number of patients using oral laxatives decreased from 16 (64%) at baseline to 11 (44%) at follow-up with SNS treatment, but this difference was not statistically significant ($P = .16$). Thirteen patients (52%) had an appendicostomy or cecostomy in place at baseline, but only 12 (48%) were regularly using ACE at baseline. At follow-up, the number of patients using ACE had decreased to five (20%, $P = .03$). Antegrade continence enema use decreased from a median of 7 (IQR 7-7) antegrade enemas per week to 0 (0-3.3, $P < .01$) at follow-up. Six of 25 patients (24%) fulfilled our criteria for successful response and were no longer receiving laxatives or ACE at follow-up. Four of these six patients had functional constipation and the remaining two patients had organic causes of constipation.

Patient-reported outcomes

Table 1 compares the three patient-reported measures of symptom severity and quality of life at baseline and follow-up after SNS treatment. Scores were recorded at a median of 1.7 years (IQR 1.4-2.3) after SNS initiation. Three patients (12%) were missing baseline scores and two patients (8%) were missing the FISI score at follow-up. The total GSS score improved significantly ($P = .01$), and the constipation, abdominal pain, and abdominal discomfort components of the GSS ($P < .01$, $P = .02$, $P = .01$ respectively). The Lifestyle, Coping/Behavior, and Embarrassment domains of the FiQL improved significantly ($P < .01$, $P < .01$, $P < .01$ respectively). The FISI improved significantly as well ($P < .01$).

Complications

Six patients (24%) had complications from SNS requiring further surgery. Four patients developed local infection of the subcutaneous pocket created for implantation of the pulse generator. All four patients underwent removal and subsequent replacement of SNS. One patient developed a second infection requiring a second SNS removal. In this patient, SNS was not replaced after it was removed for the second time.

Two patients developed displacement or malfunction of the SNS lead and required lead revision. The first of these patients experienced lead displacement after the first stage procedure but prior to permanent implantation of the pulse generator during the second stage. This patient therefore needed a second procedure to reposition the lead, after which the patient experienced improvement in both constipation and urinary symptoms. This patient then underwent a third procedure for permanent implantation of the pulse generator. The second patient who required lead revision had already undergone SNS removal and replacement because of a prior wound infection. Approximately 12 months
after SNS replacement, the patient developed both worsened constipation and nocturnal enuresis and was found to have a malfunctioning lead. She underwent lead revision with subsequent symptomatic improvement.

### TABLE 1: Clinical symptoms and patient-reported outcomes at baseline and follow-up after SNS treatment.

<table>
<thead>
<tr>
<th>Outcome</th>
<th>Baseline</th>
<th>Follow-up</th>
<th>P-value</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Clinical Symptoms, n (%)</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Defecation &lt;3 times per weeka</td>
<td>4/22 (18%)</td>
<td>3/22 (14%)</td>
<td>1.00</td>
</tr>
<tr>
<td>Fecal incontinence</td>
<td>18/25 (72%)</td>
<td>5/25 (20%)</td>
<td>&lt;.01</td>
</tr>
<tr>
<td>Urinary incontinence</td>
<td>14/25 (56%)</td>
<td>7/25 (28%)</td>
<td>.04</td>
</tr>
<tr>
<td>Abdominal paina</td>
<td>15/22 (68%)</td>
<td>9/22 (41%)</td>
<td>.11</td>
</tr>
<tr>
<td><strong>Patient-Reported Outcomes, median (IQR)</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>GSS</td>
<td>59.7 (42.4-72.2)</td>
<td>80.6 (55.6-88.9)</td>
<td>.01</td>
</tr>
<tr>
<td>FIQL: Lifestyle</td>
<td>3.0 (2.3-3.9)</td>
<td>3.9 (3.3-4.0)</td>
<td>.01</td>
</tr>
<tr>
<td>FIQL: Coping/Behavior</td>
<td>2.8 (2.2-3.9)</td>
<td>3.7 (3.3-4.0)</td>
<td>&lt;.01</td>
</tr>
<tr>
<td>FIQL: Depression</td>
<td>2.8 (2.4-3.6)</td>
<td>3.3 (2.9-3.6)</td>
<td>.08</td>
</tr>
<tr>
<td>FIQL: Embarrassment</td>
<td>3.0 (1.7-3.3)</td>
<td>3.3 (2.3-4.0)</td>
<td>.01</td>
</tr>
<tr>
<td>FISI</td>
<td>32.5 (26.0-39.0)</td>
<td>30.0 (12.0-40.0)</td>
<td>.01</td>
</tr>
</tbody>
</table>

- Baseline defecation frequency and abdominal pain was unavailable for three patients.
- GSS, PedsQL Gastrointestinal Symptom Scale; higher scores suggest improvement; FIQL, Fecal Incontinence Quality of Life Scale; higher scores suggest improvement; FISI, Fecal Incontinence Severity Index; lower scores suggest improvement.

Shortly after the second stage procedure, one patient experienced lower extremity numbness and discomfort with sitting. This patient underwent repositioning of the SNS pulse generator within a month of the second stage procedure with subsequent resolution of the numbness and discomfort.

One patient experienced a relapse of symptoms requiring further surgical intervention. After a brief period of improvement in constipation symptoms for the three months following SNS initiation, this patient’s symptoms gradually returned over the following year despite SNS treatment. This patient subsequently underwent partial colectomy with colorectal anastomosis approximately 15 months after SNS placement. This patient continues to have an SNS in place but has had infrequent stools and regular fecal incontinence at follow-up.
Chapter 15

**Patient benefit and parent satisfaction**

We called families a median of 2.3 years (IQR 2.1-2.7) after SNS initiation. We were able to contact the parents of 17 patients, including all six of the children who experienced complications after SNS initiation requiring further surgery. Sixteen parents (64%) agreed to participate and completed the GCBI and satisfaction questionnaire. The patient who declined to participate was one of the children who had experienced a local infection after SNS initiation. Median GCBI was +42.7 and 15 parents (94%) reported GCBI scores >0, indicating positive health-related benefit. The patient who had a negative GCBI score had undergone SNS replacement secondary to wound infection. Fourteen parents (88%) would proceed with SNS if given the opportunity to remake their decision. Two parents (13%) would not proceed with SNS, one because of complications requiring further surgery and the other because of lack of improvement. All 16 parents would recommend SNS to others. A summary of parent responses when asked to explain their answers to our satisfaction questionnaire is presented in Table 2. A full description of parent responses is provided in Supplemental file 1.

<table>
<thead>
<tr>
<th>Positive responses (n)</th>
<th>Negative responses (n)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. SNS decreased the need for antegrade continence enemas (4)</td>
<td>7. SNS improved urinary symptoms but not constipation symptoms (3)</td>
</tr>
<tr>
<td>2. SNS decreased the need for oral laxative treatment or bowel clean outs (3)</td>
<td>8. Process of starting SNS and adjusting settings took too long (3)</td>
</tr>
<tr>
<td>3. SNS improved constipation symptoms (3)</td>
<td>9. SNS did not help symptoms (1)</td>
</tr>
<tr>
<td>4. Parents have already recommended SNS to others (3)</td>
<td>10. SNS led to complications (1)</td>
</tr>
<tr>
<td>5. SNS was life-changing (2)</td>
<td></td>
</tr>
<tr>
<td>6. SNS improved quality of life (2)</td>
<td></td>
</tr>
</tbody>
</table>

Three parents reported that SNS improved urinary symptoms but not constipation symptoms, raising the possibility that parents of children with urinary symptoms reported greater patient benefit or parent satisfaction because of improvement in urinary symptoms alone. When we divided the 16 responding parents into parents of children with and without urinary symptoms, we found that the 12 parents of children with urinary symptoms...
Outcomes of sacral nerve stimulation

actually reported a lower median GCBI (+31.25) than the four parents of children without urinary symptoms (+61.46). Ten of the 12 parents (83%) of children with urinary symptoms would proceed with SNS if given the opportunity to remake their decision, while all of the parents of children without urinary symptoms would proceed with SNS.

Analysis of patients with functional constipation

Sixteen of our 25 patients (64%) had functional constipation according to Rome III criteria (63% female, median age at SNS initiation 11 years, range 6-19 years). At the time of data collection, patients with functional constipation had been treated with SNS for a median of 2.5 years (IQR 2.1-2.7). At baseline, 11 patients (69%) had concurrent fecal incontinence, 12 patients (75%) had urinary incontinence, and one (6%) had urinary retention.

Clinical symptoms and constipation treatment at baseline and follow-up for patients with functional constipation after SNS treatment are shown in Table 3. We found significant decreases in the presence of fecal incontinence, urinary incontinence, and abdominal pain at follow-up. The number of patients with functional constipation who were using oral laxatives and antegrade continence enemas decreased at follow-up, but these differences were not statistically significant. Ten of the 16 patients (63%) with functional constipation fulfilled our criteria for successful response at follow-up. Four of 16 patients (25%) fulfilled our criteria and were no longer receiving laxatives or ACE at follow-up. Four of 16 patients (25%) experienced complications requiring further surgery.

<table>
<thead>
<tr>
<th>Outcome</th>
<th>Baseline</th>
<th>Follow-up</th>
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<tr>
<td>Clinical symptoms, n (%)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Defecation &lt;3 times per week&lt;sup&gt;a&lt;/sup&gt;</td>
<td>3/14 (21%)</td>
<td>3/14 (21%)</td>
<td>1.00</td>
</tr>
<tr>
<td>Fecal incontinence</td>
<td>11/16 (69%)</td>
<td>3/16 (19%)</td>
<td>&lt;.01</td>
</tr>
<tr>
<td>Urinary incontinence</td>
<td>13/16 (81%)</td>
<td>6/16 (38%)</td>
<td>.01</td>
</tr>
<tr>
<td>Abdominal pain&lt;sup&gt;a&lt;/sup&gt;</td>
<td>12/14 (86%)</td>
<td>6/14 (43%)</td>
<td>.02</td>
</tr>
<tr>
<td>Constipation treatment, n (%)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Using oral laxatives</td>
<td>11/16 (69%)</td>
<td>7/16 (44%)</td>
<td>.15</td>
</tr>
<tr>
<td>Using antegrade continence enemas</td>
<td>6/16 (38%)</td>
<td>3/16 (19%)</td>
<td>.24</td>
</tr>
</tbody>
</table>

<sup>a</sup>Baseline defecation frequency and abdominal pain was unavailable for two patients.
Twelve of 16 parents (75%) of patients with functional constipation provided information on patient benefit and parent satisfaction. The median GCBI score for patients with functional constipation was +39.58. Eleven of 12 parents (92%) reported that they would proceed with SNS if given the chance to remake their decision.

**Presence of concurrent fecal incontinence**

Eighteen of our 25 patients (72%) had constipation with concurrent fecal incontinence and seven patients (28%) had constipation alone. We did not find significant differences in ability to discontinue laxative or ACE use after SNS treatment between patients with and without concurrent fecal incontinence. We also did not find any differences between the GCBI scores or satisfaction questionnaire responses of the two groups. Interestingly, patients without concurrent fecal incontinence required further surgery after SNS more often (4/7, 57%) than patients with concurrent fecal incontinence (3/18, 17%; \( P = .04 \)).

**DISCUSSION**

Our study suggests that SNS can be an effective, long-term treatment option for children with refractory constipation. We found that SNS led to continued improvement in both symptoms and quality of life at 2 years after treatment initiation, with particular improvement in concurrent fecal incontinence. At follow-up, 68% of our patients fulfilled our criteria for successful response and 24% fulfilled criteria without concurrent laxative or ACE use. Although a quarter of patients developed a complication requiring additional surgery, nearly all parents reported health-related benefit from the treatment and all parents would recommend SNS treatment to other families with children experiencing similar symptoms.

Although long-term outcomes of SNS treatment for adults with defecation disorders have been reported, information on long-term outcomes for children with defecation disorders remains limited. In the first study of SNS treatment for children with constipation, van Wunnik et al. described symptomatic improvement in 12 adolescent females with functional constipation at up to 12 months of follow-up.\(^9\) Sulkowski et al. reported short-term outcomes of the first 29 children treated with SNS at our institution, which included 22 children who had constipation at baseline. This group, which included both children with organic causes of constipation and functional constipation, experienced significant improvement both symptomatically and in quality of life at a median of 4-5 months after SNS initiation.\(^10\) A portion of this group was included in the current study. Van der Wilt et al. recently published a follow-up study that included the group originally described by van Wunnik, and they reported significant symptomatic improvement in 27 female children and adolescents who had been treated with SNS for 12-37 months. The authors concluded that
although response to SNS was heterogeneous in their cohort, SNS does appear to provide sustained benefit for children with functional constipation refractory to conventional treatment.\textsuperscript{11} Our findings add to the evidence that SNS can be an effective and durable treatment option for children with refractory constipation. Although our study included both patients with organic causes of constipation and functional constipation, subgroup analysis of patients with functional constipation demonstrated clinical improvement and patient benefit that was similar to that of our entire cohort.

However, it is important to recognize that although SNS has become an established treatment option for adults with refractory fecal incontinence, evidence for its use for adults with constipation is less clear.\textsuperscript{6} In fact, two recent randomized crossover studies did not find SNS to be more effective than sham stimulation for adults with refractory constipation.\textsuperscript{7,8} In children, fecal incontinence often occurs secondary to poorly controlled constipation and is commonly used as a primary outcome to define treatment success in studies of pediatric constipation.\textsuperscript{19,20} The majority of the patients in our study suffered from concurrent fecal incontinence prior to initiation of SNS, and we did find a significant decrease in the number of children suffering from fecal incontinence after SNS treatment. This improvement was also reflected by corresponding changes in FIQL and FISI scores.

Therefore, the question arises whether the clinical improvement demonstrated in our study was the result of improvement in fecal incontinence alone rather than in the child’s underlying constipation. Although our analysis is limited by our sample size, our comparison of outcomes between patients with and without concurrent fecal incontinence suggests that SNS can be beneficial for children with constipation regardless of the presence of fecal incontinence. However, particularly given recent findings in the adult literature, this is a question that needs to be evaluated further as we continue to search for predictors of response to SNS treatment.

Families generally viewed SNS treatment as beneficial to their children. The median GCBI score of our sample was +42.7, which suggests a strongly positive perceived benefit after SNS treatment initiation. This score was higher than the reported GCBI of a number of established surgical procedures, including tonsillectomy and adenoidectomy (+21), otoplasty (+24.4), and seoptoplasty (+35.1).\textsuperscript{21-23} Comparable measures of patient benefit are not available for other treatment options for pediatric intractable constipation, such as ACE or colonic resection.

As part of our assessment of parent satisfaction, we asked parents to explain their answers to our satisfaction questionnaire without further prompting. A theme we noticed in their responses was that SNS allowed their children to live a more “normal” life when compared
to ACE treatment. One parent reported that before starting SNS treatment, their family was confined to their home every evening because of their child’s daily ACE regimen, which would often take an hour or more to complete. After starting SNS treatment, their child was able to decrease and ultimately discontinue ACE use, allowing the family to leave the home without worrying about setting time aside for ACE administration. This parent’s experience is supported by our recent findings that SNS can allow decreased ACE use in children with refractory constipation already treated with ACE.24

A number of parents had negative comments regarding SNS treatment. A few parents felt that the length of time involved in the evaluation prior to starting SNS treatment, the two-stage initiation process, and waiting for symptomatic improvement was longer than anticipated. A handful of parents also noted that urinary symptoms improved more than constipation or fecal incontinence, which likely contributed to their perception of benefit from the treatment.

It is concerning that a quarter of our patients experienced complications that required further surgery and that more than one surgery was often required. For example, each of our patients who developed a wound infection underwent two operations, the first for device removal and the second for replacement. One of these patients went on to develop a second infection and another required surgical lead revision. In their series of 27 children, van der Wilt et al. reported that 12 children required further surgery, but these were primarily for repositioning of the lead or stimulator. Only one patient developed an infection.11 Our experience was consistent with what has been reported in adult studies. In a recent review of nearly 2000 patients reported in the literature to have undergone SNS for fecal incontinence, Bielefeldt reported that 18.6% of patients experienced complications requiring further surgery. Device removal was generally secondary to infection.25 The complication rate found in our sample emphasizes the need for further studies evaluating for predictors of complication development in our patient population. The comparative evaluation of less-invasive neurostimulation techniques may be useful in identifying safer alternative treatments.

Our study has several limitations. The observational nature of our study and the lack of a control or comparison group is a weakness that we will address in future studies. We recently began a study comparing the outcomes of children with refractory constipation treated with various treatment strategies. Although our study includes one of the larger cohorts of children with constipation treated with SNS in the literature, our sample size remains limited and prevents us from performing further subgroup analysis that could allow identification of patient characteristics associated with SNS response or complications. We were also able to reach only 17 of 25 parents during our assessment of patient benefit and parent
satisfaction, which may have resulted in bias. However, the 16 parents who completed our interview included 5 of the 6 children who experienced complications requiring further surgery, and complications were the primary concern raised by the parents who completed our interview.

In conclusion, we found that SNS can lead to durable improvement in both symptoms and quality of life in children with constipation treated with SNS. Nearly all parents reported benefit from the treatment and all would recommend the treatment to others. However, a quarter of our patients experienced complications from SNS requiring further surgery. Further studies evaluating for predictors of both treatment response and complication development are needed before there is more widespread acceptance of this novel treatment modality.
REFERENCES

SUPPLEMENTAL FILE 1. Parent responses from all 16 participants when asked to explain their answers to our satisfaction questionnaire. Responses are followed by numbers corresponding to summary statements provided in Table 2.

SNS was life-changing for our family. Washouts were difficult, prevented travel, would take one to one and a half hours to complete, and bowel movements could still occur in the middle of the night. (1, 5, 6)

SNS helped with bowel symptoms. It works as long as he is compliant with medications. I would absolutely recommend SNS to others. (3)

SNS helped symptoms. (3)

He wanted SNS because he didn’t like his flushes. (1)

SNS helped bladder symptoms, but there was no change in bowel symptoms. (7)

Bladder now continent, but no bowel change. Bladder symptoms resolved and with very few side effects. Infection was an anomaly. Process took too long. (7, 8)

Starting SNS was a long process but worth it. It’s great. (8)

She expels the SNS device. She has had lots of complications. Her SNS has been removed. (10)

Quality of life dramatically improved after SNS. Quick effect. ACE reversed. We have already recommended SNS to others. (1, 4, 6)

Urinary problems are much better. Bowel problems are better after colon resection. Initially there was no difference after SNS and the search for the right settings took a long time. (7, 8)

I’m not sure how much SNS has helped her. That being said, she has oppositional defiant disorder and will not take her medications a lot of the time. (9)

SNS allows for hope of a normal life, unlike a cecostomy tube or colostomy. She has already recommended SNS to others. She was one of the first to have it done and spoke to others about it. (1, 4)

SNS has been the only thing that has helped. We had been told her symptoms were a failure on our part as patients. She had tried bladder training and mineral oil, bisacodyl, and Miralax. She used to have bowel movements only with suppositories. We were told there would be a referral to Children’s Services. She has had multiple invasive procedures previously. It has changed her life completely. She wishes she had gotten this before being tried on all these medicines. We would suggest this before going through so many different medications. (2, 3, 5)

Before SNS, he had to be hospitalized every 4-6 weeks for NG tube cleanouts. (2)

His medications and enemas have changed. Now, it’s great. He was previously taking 5 mg of bisacodyl 8 times per day. The only downside of SNS is that he can’t do deadlifts, which he says is probably not good for his body anyway. It feels as though his mood is better. He can eat better and keep weight on, and he no longer has to take as many medications. (2)

She was still in pull-ups at age 7. Now she has full control of her bladder, day and night, although not her bowels. We have referred another patient. (4)