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Sacral nerve stimulation in the treatment of bowel dysfunction from imperforate anus: A case report



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ABSTRACT

INTRODUCTION: Bowel dysfunction (fecal incontinence and constipation) presents in over 50% of patients after treatment of congenital anal malformations. Sacral nerve stimulation (SNS) for the treatment of fecal incontinence improves function in the majority of patients. We present a case report of the treatment of bowel dysfunction with sacral nerve stimulation in a patient with a history of an imperforate anus.

PRESENTATION OF CASE: A twenty year-old female with a history of imperforate anus at birth, repaired during infancy with anorectoplasty, presented with fecal incontinence and constipation. Since childhood, she had been suffering from intermittent constipation with worsening fecal incontinence in early adulthood. Examination revealed mild anal stenosis and mucosal prolapse. Endoanal ultrasound demonstrated intact internal and external sphincter with low resting and squeeze pressures on anal manometry. Flexible sigmoidoscopy was normal. The patient underwent permanent sacral nerve stimulation with a primary goal of improvement in continence and, secondarily, for the alleviation of intermittent chronic constipation.

DISCUSSION: At 15 month follow-up, the patient had improvement in fecal incontinence (CCIS of 14 pre-SNS to 1 post-SNS), constipation (CCCS of 28 pre-SNS to 20 post-SNS), and quality of life (FIQOL improved in lifestyle (3.7), coping/behavior (3.4), self perception (3.9), and social embarrassment (4.5).

CONCLUSION: Sacral nerve stimulation for the treatment of bowel dysfunction in adults secondary to imperforate anus can be performed safely and with good results.

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1. Introduction

Bowel dysfunction affects a significant portion of patients with a history of congenital anal malformations, with incontinence and constipation being the most frequent symptoms [1]. Sacral nerve stimulation (SNS) improves function in a majority of patients with fecal incontinence [2,3]. Although results of SNS treatment for constipation are not as robust, they are, nevertheless, promising [4]. To our knowledge, the treatment of bowel dysfunction with sacral nerve stimulation in patients with a history of congenital anal anomaly has been documented in a total of 5 patients, with 4 of them proceeding to permanent implantation [5–7]. Improvement in continence and/or constipation was documented in 4 of the permanently implanted patients. We present our experience in the treatment of bowel dysfunction with permanent sacral nerve

stimulation in a patient with a history of low congenital anal malformation. The present study has been developed with the standards established by the CARE criteria in the documentation of case reports [8].

2. Presentation of case

A twenty year-old female presented to our clinic with complaints of worsening fecal incontinence coupled with a history of intermittent constipation. She had a history of a low imperforate anus and was treated in infancy, with an anorectoplasty, or pull through procedure. She did not have vertebral, cardiac, trachea-esophageal, renal, or limb defects (VACTERL Association). From the age of 12, she had suffered from persistent constipation requiring digitalization, frequent enema usage and oral laxative administration (Fig. 1). Her Cleveland Clinic Constipation Score (CCCS) was 28. By the age of twenty, her bowel dysfunction continued to evolve as fecal incontinence became her dominant symptom in the background of intermittent constipation manifested by frequent soiling and bowel accidents with a Cleveland Clinic Incontinence Score

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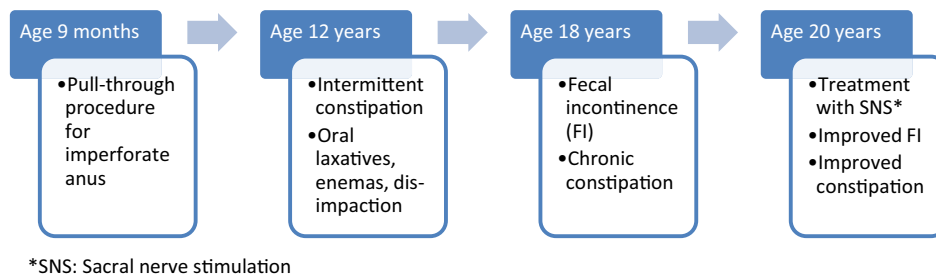


Fig. 1. Timeline of patient's interventions, presentations, and treatments.

(CCIS) of 14. On exam she had a patulous anus with mild anal stenosis and mucosal prolapse with poor resting/squeeze tone along with a thin perineal body. Anal manometry revealed low resting (29.6 mmhg) and squeeze (60.3 mmhg) pressures, along with low compliance. Endoanal ultrasound showed a normal puborectalis sling with an attenuated but intact internal sphincter and external sphincter. Flexible sigmoidoscopy was significant for mild anal stenosis; otherwise the descending colon, sigmoid and rectum were normal. The patient's incontinence was her most significant problem as it was negatively affecting her quality of life. Her pre-SNS Fecal Incontinence Quality of Life (FIQOL) scale was most notable in regards to lifestyle alteration (1.1), poor coping and behavior (1), along with depression and poor self perception (2.5). Although weak, her sphincters were intact, and thus she was not a candidate for sphincteroplasty. Therefore we offered her sacral nerve stimulation for the primary treatment of incontinence with a secondary goal of treating her constipation.

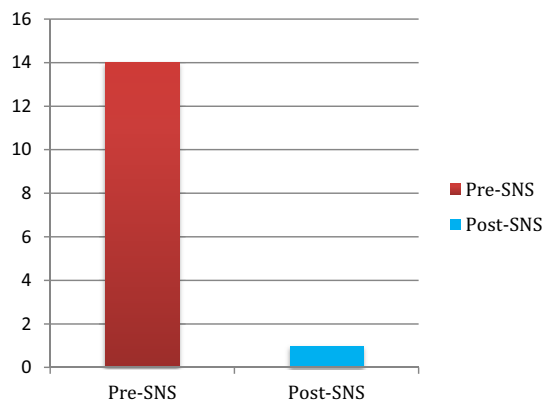
3. Intervention and results

The patient underwent test stimulation with SNS involving the percutaneous placement of a temporary electrode targeted toward the third sacral nerve aided by fluoroscopic guidance. The temporary lead was connected to an external stimulator that provided pulse electrical stimulation to the sacral nerve. After two weeks, the patient had a greater than 50% improvement in fecal continence as documented with a bowel dysfunction diary. We proceeded with permanent implantation of the electrical stimulator subcutaneously overlying her right gluteus muscle 2 weeks later. Postoperatively she maintained improvement in fecal incontinence with CCIS decreasing from 14 pre-SNS to 1 post-SNS at 15 month follow up (Fig. 2). In addition, constipation improved marginally with CCCS of 28 pre-SNS to 20 post-SNS (Fig. 3). She had reported a significant improvement in her quality of life as her FIQOL improved in the areas of lifestyle (3.7), coping/behavior (3.4), self perception (3.9), and social embarrassment (4.5) (Fig. 4).

4. Discussion

Congenital anal malformations are frequently associated with bowel dysfunction in adulthood. At least one-third of patients report long term functional problems related to constipation and soiling [9]. Sacral nerve stimulation is effective in the treatment of fecal incontinence in the majority of patients [2,3]. With the initial introduction of sacral nerve stimulation by Matzel et al. [10] for fecal incontinence, the indications were relatively narrow, excluding patients with questionable sphincter defects, neurogenic incontinence, and surgical incontinence. However, over the past 20 years, the indications for sacral nerve stimulation have continuously expanded with application to those previously excluded, with encouraging results.

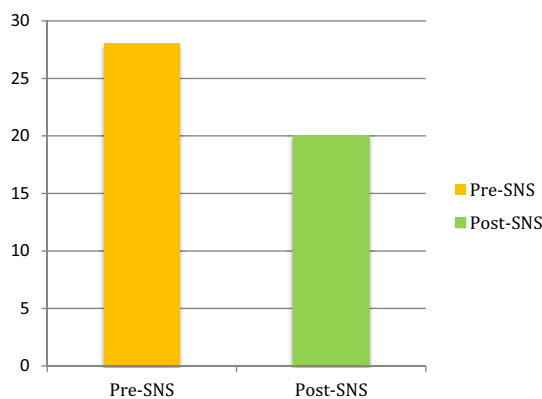
Cleveland Clinic Incontinence Score



*SNS: Sacral nerve stimulation

Fig. 2. Cleveland Clinic Incontinence Score (CCIS) before and after treatment.

Cleveland Clinic Constipation Score



*SNS: Sacral nerve stimulator

Fig. 3. Cleveland Clinic Constipation Score (CCCS) before and after treatment.

The use of sacral nerve stimulation in adults with bowel dysfunction and a history of congenital imperforate anus is a relatively recent application. Since 2013, there have been European reports of SNS treatment for adults with bowel dysfunction and a history of congenital anal malformation. A total of 5 patients were identified in the literature with varying VACTERL association (sacral agenesis), fecal incontinence, and constipation [5–7] who had undergone sacral nerve stimulation with improvement in bowel dysfunction. SNS has been previously applied in children with bowel dysfunction

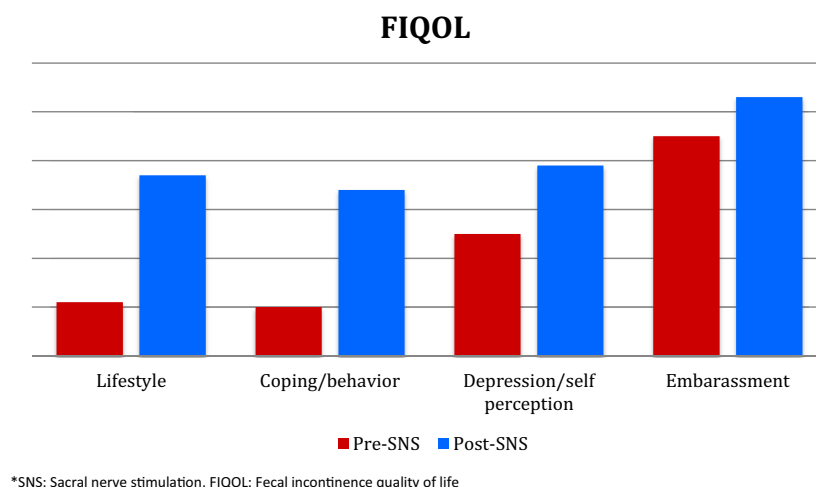


Fig. 4. Fecal Incontinence Quality of Life (FIQOL) scale pre and post treatment. Scale ranges from 1 to 5, with 1 indicating a lower functional quality of life.

tion secondary to imperforate anus, with improvement in FIQOL and FISI scores [11].

Our patient did not have a VACTERL association with her congenital anal malformation and did not suffer from sacral agenesis. SNS had improved the patient's fecal incontinence, quantified by an improvement in her CCIS, along with a subjective feeling of increased control over her urge to defecate. There was a modest improvement in her constipation. As SNS has been shown to improve constipation [12], there are inconsistent reports of the continued usage of laxatives in patients despite an improvement in CCCS which is not clearly understood. Our patient continued to use oral laxatives; however she had decreased administration of enemas and digitalization with overall more frequent defecation. The complex and evolving bowel dysfunction symptoms in our patient showed improvement along with an important enhancement in her quality of life, which was uniquely documented in the present case report with the validated FIQOL scale [13].

To our knowledge, this is the first case report of SNS for bowel dysfunction in an adult with a history of congenital anal malformation in the United States. As the indications for SNS have been expanding for the past 20 years, this report further supports the use of SNS in the relatively new indication of bowel dysfunction in patients with a history of congenital anal malformations. In addition, our unique use of a validated quality of life instrument underscores not only improvement in function, but also the psychosocial well-being in these patients.

The present report outlines the following learning points. First, bowel dysfunction presents with complex and often paradoxical symptoms in patients with a history of congenital anal malformation repaired in infancy. Fecal incontinence often becomes the dominant symptom in adulthood and has a detrimental effect on quality of life. SNS can be used to treat fecal incontinence and may improve other accompanying symptoms of bowel dysfunction such as urgency and constipation. Secondary to the history of congenital anal malformation, the practitioner must be aware of possible sacral agenesis as this can complicate the placement of stimulator leads in the foramina of the sacrum.

5. Conclusion

Sacral nerve stimulation can improve bowel dysfunction and quality of life in adult patients with a history of congenital anal malformation. As these results are encouraging, there is a need for adequately powered studies to examine and potentially validate SNS application in this specific population.

Sources of funding

None.

Consent

Appropriate consent from the patient was obtained per institutional protocol and guidelines. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Conflict of interest

The authors report that there are no conflicts of interest.

Ethical approval

Advocate Health Care Institutional Review Board does not require review for case reports.

Author contributions

Saleh M. Eftaiha, MD: drafting of the article, critical revision of the article for important intellectual clinical content; (2) George Melich, MD: equal contribution as lead author, drafting of the article, revision of the article for important intellectual clinical content; (3) Ajit Pai, MD: revision of the clinical and intellectual content of the article; (4) Slawomir Marecik, MD: revision of the clinical and intellectual content of the article; (5) Leela M Prasad, MD: revision of the clinical and intellectual content of the article; (6) John J Park, MD: revision of the clinical and intellectual content of the article.

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John Park, MD.

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