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ORIGINAL ARTICLE

Systematic review: sacral nerve stimulation in the treatment of constipation and fecal incontinence in children with emphasis in anorectal malformation

Lindel Dewberry[2] · Andrew Trecartin[1] · Alberto Peña[1] · Marie St. Pierre[3] · Andrea Bischoff[1]

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Abstract

Background Sacral nerve stimulation (SNS) is frequently used for constipation and fecal incontinence in the adult literature. The purpose of this study is to perform a systemic review of the literature for SNS for constipation and fecal incontinence in children with emphasis in anorectal malformations.

Methods Systematic literature review was conducted to include all SNS studies in patients < 19 years of age. Studies were separated into those for (1) constipation, (2) bowel and bladder dysfunction, and (3) anorectal malformations.

Results 28 articles were included in the review: (1) 12 constipation (269 patients) and (2) 16 bowel and bladder dysfunction (441 patients). Some studies overlapped groups, as they included some patients with anorectal malformations (4 articles and 29 patients). Constipation studies included slow transit and retention constipation and showed varying degrees of improvement. For bowel and bladder dysfunction, studies also reported varying degrees of improvement using different measures (number of bowel movements per day, transit times, and soiling improvement). There was no specific description of the results in anorectal malformation (ARM) cases and also information regarding specific ARM type, sacral ratio, or presence of tethered cord.

Conclusions SNS for constipation and urinary problems seems to be promising. Data are limited and heterogeneous, and SNS cannot be definitively encouraged or discouraged in patients with ARM, based on current studies. Future studies should include more objective measurements of bowel outcomes and specify outcomes related to patients with anorectal malformations including information regarding their specific malformation, sacral ratio, and presence of tethered cord. Complications' rate is considerable high.

Keywords Sacral nerve stimulation · Anorectal malformation · Constipation · Incontinence

Introduction

Invasive and noninvasive sacral nerve stimulation (SNS) have been used for a variety of indications in the adult population including urinary incontinence, overactive bladder, dysfunctional elimination syndrome, slow colonic transit, constipation, and fecal incontinence [1–7]. The experience of this type of treatment in pediatrics is rather limited.

In regard to noninvasive sacral nerve stimulation, there are several different approaches including transcutaneous electrical stimulation using interferential current (TISEC), transcutaneous electrical nerve stimulation (TENS), and transcutaneous posterior tibial nerve stimulation.

The purpose of this study to perform a systematic review of SNS for constipation and fecal incontinence in children, with and without anorectal malformation (ARM).

✉ Andrea Bischoff
andrea.bischoff@childrenscolorado.org

[1] Department of Pediatric Surgery, International Center for Colorectal and Urogenital Care, Children's Hospital Colorado, University of Colorado, Box 323, 13123 East 16th Avenue, Anschutz Medical Campus, Aurora, CO 80045, USA

[2] Department of Surgery, University of Colorado, Aurora, CO, USA

[3] Medical Library, Children's Hospital Colorado, Aurora, CO, USA

Methods

Study design

A systematic literature review was conducted of all studies regarding sacral nerve modulation in the pediatric population (< 19 years of age) from 2010 to 2018. PubMed was used to identify appropriate articles with the following MESH terms: "fecal incontinence," "constipation," and "transcutaneous electric nerve stimulation." The following search strategy was used: (((((((“anorectal malformation”) OR “colorectal malformations”) AND (“2010/01/01”[PDat]: “2018/12/31”[PDat]))) OR (((“Fecal Incontinence”[Mesh]) AND “Constipation”[Mesh]) AND (incontinence OR “fecal incontinence” OR constipation))) AND (“2010/01/01”[PDat]: “2018/12/31”[PDat]))) AND ((“interstim therapy”) OR (((((((nerve stimulation) OR “Transcutaneous Electric Nerve Stimulation”[Mesh]) OR “sacral nerve stimulation”) OR “sacral neuromodulation”) OR “spinal nerve root”) OR “nerve stimulation”) OR neuromodulation)). This resulted in 157 publications. The exclusion criteria included, publications in languages other than English, adult series, sacral nerve modulation used in cases of urologic problems, involved non-human subjects, and single case reports. After exclusion, we were left with 28 articles spanning from 2008 to 2018.

We decided to separate the articles into three categories: (A) those who were dealing with the treatment of constipation only (12 articles, 269 patients) and (B) Bowel and Bladder Dysfunction (16 articles, 441 patients). We could not find an article describing the use of sacral nerve stimulation specifically for patients with anorectal malformations. However, within groups A and B, some authors included cases with anorectal malformations, so we included those cases in group C (29 cases).

Statistical analysis

Statistical analysis could not be performed due to the heterogeneity of the data.

Results

Group A (constipation): sacral nerve stimulation for the treatment of constipation only

Selected articles included a mixture of invasive and noninvasive SNS. Nine articles [8–15] referred to the treatment of “Slow Transit Constipation” (259 cases). One article [11] was related to the treatment of “Retention Constipation” (10 cases).

Three articles [10, 16, 17] reported an improvement in 50 to 73% of the cases, consisting in the acceleration of the transit time, as measured by nuclear transit studies. Nine articles [8–15] reported different degrees of improvement as demonstrated by the increase of the number of bowel movements or decreased use of laxatives. Complications requiring device revision or removal ranged from 23 to 44%.

GROUP B (bowel and bladder dysfunction)

Sixteen articles (included both invasive and noninvasive SNS) discussed the treatment of bowel and bladder dysfunction with SNS in a total of 441 patients. In this group, the results were more difficult to interpret, since the “improvements” were frequently reported without specification as to whether or not the authors were referring to bladder function or bowel function. All studies reported different degrees of “improvement”, measured by the acceleration of the transit times, the number of bowel movements per week, or by a decrease in the number of times that some patients used their antegrade enema mechanism. In one study, 50% of patients no longer needed enemas. In another study, antegrade enema use decreased from 7 times to 1 time per week. The percentage of cases who responded to the treatment varied between 50 and 73% of the cases [1–7, 18–26] with a 6–34% complication rate requiring device revision or removal.

GROUP C (anorectal malformations)

These cases were not reported as separate articles but rather mentioned within a larger group of patients (DiLorenzo 6 cases, Sulkowski 8 cases, Lecompte 4 cases and Lu 11 cases) [4, 5, 7, 20]. It is important to mention that with the exemption of the four cases reported by Lecompte, the other three publications originated from the same institution, and therefore, we do not know if they reported the same cases in three different publications. The authors did not mention the characteristics and diagnosis of those patients who responded and those who did not respond to treatment. In addition, the specific type of anorectal malformation, characteristics of the sacrum (sacral ratio), and the presence or absence of tethered cord were not discussed in relation to outcomes. The average percentage of complications which required the removal or revision of the device was 25%. The longest follow-up was 36 months.

The study by Lecompte examined transcutaneous electrical stimulation for fecal incontinence in four patients with anorectal malformations. These ARM patients had “high” malformations and three patients had “partial” sacral agenesis. They included patients who had undergone 2 years of therapy using transit regulators, anticholinergics if needed,

antegrade enemas, transanal irrigations, abdominal and perineal rehab, and psychological support. All continued enemas throughout treatment. They performed posterior tibial nerve stimulation 20 min daily for 6 months at a frequency of 10 Hz. Their outcome measure was number of bowel movements, episodes of soiling, and the Jorge Wexner fecal incontinence score. The defecation diary demonstrated that 2/4 ARM patient's incontinence resolved, and their Wexner score decreased from 13.5 to 5. 75% stopped using antegrade enemas, 50% had spontaneous defecation, but all were still using transanal irrigation.

The other studies examined invasive SNS. The study by Sulkowski et al. included eight patients with imperforate anus without discussion of the quality of the sacrum. Of these, one patient had a myelomeningocele and another had tethered cord. Overflow incontinence with constipation versus fecal incontinence was not clearly described. In the ARM patient population, 88% reported improvement in cecostomy tube flushes and 50% reported symptomatic improvement. In terms of other measures, the FIQL domain of embarrassment improved from 2.8 to 4, Fecal Incontinence Severity Index (FISI) increased from 15.5 to 17, PedsQL GSS decreased from 11 to 8, and DES score remained unchanged at 8. The reoperation for the overall cohort was 17% [7]. The study by DiLorenzo et al. includes 6 ARM patients with constipation with soiling. Their study did not describe the type of ARM or quality of the sacrum. In the total cohort (including non ARM patients), there was a 68% improvement in constipation. The study by Lu et al. included 11 patients with ARM. Of these patients, 7 had an "abnormal sacrum" and 4 had a tethered cord. Antegrade continence enema (ACE) reduction was decreased from seven times per week to one time per week. In the overall cohort, there were six complications that required reoperation [4].

Discussion

Both invasive and noninvasive sacral nerve stimulation (SNS) appear to be a promising treatment for both, "slow transit" as well as "retention" constipation in children. Currently, the use of SNS in children is indicated for those with "intractable" constipation as an alternative or in conjunction with the use of ACE and/or colonic resection. We could not find in any of the publications a specific description of what "intractable" means. It is not unusual for clinicians to find that a patient considered "intractable" is actually manageable with a more aggressive treatment. The mean complication rate of 25% is a limitation to the use of this technology.

In the anorectal malformation population, there are multiple factors that influence continence outcomes, and these need to be taken into account when considering therapeutic options, including SNS. The first consideration is the type of anorectal malformation. More complex anorectal malformations (i.e., rectourethral and rectobladderneck) have high rates of fecal incontinence. Current studies do not specify type of ARM. The evaluation of patients with ARM must include an objective assessment of the sacrum, such as sacral ratio. Terms such as "dysplastic," "hypoplastic," and "abnormal" should be eliminated. Some articles describe the sacrum in vague terms such as "partial agenesis" or "abnormal sacrum," but neither specify sacral ratio [4].

Limitations of the current studies for the use of SNS in children with constipation make it difficult to provide definitive recommendations regarding the use of this therapy in patients with anorectal malformations. Studies are missing a measurement of the magnitude of the constipation as well as the magnitude of improvement. The increase in the number of bowel movements is not an objective way to evaluate the results of SNS. It is well known by most clinicians dealing with constipated patients, that, contrary to representing an improvement of the condition, the increase in the number of bowel movements per week or per day, is frequently an ominous sign of worsening stool burden. Patients who are severely constipated may have 2, 3, or 5 small bowel movements per day and yet be fecally impacted. We have learned that constipation means incapacity to empty the rectosigmoid, frequently independent of the number of bowel movements. None of the authors mentioned an evaluation of the degree of emptiness of the rectosigmoid before and after the treatment. Other measurements including constipation scores (Rome III, Wexner [27, 28]) are also deficient, as they use non-reliable parameters such as the number of bowel movements, the "need for help."

Furthermore, studies are heterogeneous in regard to inclusion criteria ranging from constipation with overflow incontinence to true fecal incontinence. The authors frequently mixed cases with different origins and types of disease.

It is important to describe the characteristics of those cases who responded to the treatment as compared to those who did not respond, in order for clinicians to learn when this type of treatment is indicated. Finally, follow-up is limited with the maximum length of follow-up of 36 months. The long-term effects of SNS are unknown, and how long therapy needs to be continued remains unanswered.

In conclusion, there is not enough available evidence in favor or against the use of this therapeutic methodology in cases of anorectal malformations. Future studies to evaluate the value of this type of treatment must include: (1) description of the specific type of malformation, avoiding archaic, misleading terminology such as "high", "intermediate", and low"; (2) description of the characteristics of the sacrum, using objective measurements, such as the "sacral ratio" and avoiding vague description such as "hypoplastic," "malformed," or "abnormal"; and (3) the results for cases of fecal incontinence must be more specific and describe if

the patients switched from having no voluntary bowel movements to having full control with no fecal soiling. Finally, as surgeons gain more experience in invasive SNS, we hope that the number of complications decreases. A 25% risk of complications is a high proportion that limits the use of this device.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Statement of human rights All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee (include name of committee + reference number) and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was not obtained, as identifiable information was not included in the manuscript.

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