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TRANSRECTAL ELECTROSTIMULATION THERAPY FOR NEUROPATHIC BOWEL DYSFUNCTION IN CHILDREN WITH MYELOMENINGOCELE

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ABSTRACT

Purpose: We attempted to evaluate the efficacy of transrectal bowel stimulation for neurogenic bowel dysfunction in children with myelodysplasia.

Materials and Methods: Daily sessions of transrectal electrostimulation were performed on an outpatient basis for 2 to 3 weeks on children with myelodysplasia and stool incontinence. If benefits were noted, 5 to 10 additional daily sessions were performed. Complete success was defined as improvement in all parameters of interest, including decrease in the frequency of daily bowel movements, increased sensation, increased ability to hold stool and a significant subjective change in bowel habits. Moderate success implied improvement in 1 to 3 parameters and treatment failure was defined as lack of improvement in any parameter.

Results: A total of 55 children 2 to 14 years old (mean age 6.7) completed a mean of 18 daily sessions per patient of bowel electrostimulation. Followup ranged from 1 to 6 years. Diapers are no longer required due to defecation problems in 14 children older than 3 years. Complete success was achieved in 20 cases (36.3%) and moderate success in an additional 30 (54.5%, overall success rate 90.8%). Specifically, 89% of the patients reported elimination of stooling accidents, 82% reported increased sensation and 71% were able to hold the bowel movement. Overall 68% of the patients noticed significantly improved bowel function. Complete/moderate success of transrectal electrostimulation was statistically significant for all 4 parameters ($p < 0.05$), and complete success was significant for increased sensation, ability to hold and episodes of accidents. Therapy failed in 5 children (9%). There were no untoward effects.

Conclusions: Transrectal electrostimulation is a well tolerated and minimally invasive modality that provides sustainable improvement in stool continence in children with myelomeningocele and neuropathic bowel dysfunction.

KEY WORDS: meningomyelocele, electric stimulation, fecal incontinence

Neural tube defects, particularly myelomeningocele, occur in 1 to 4/1,000 live births.[1] Of the myriad of clinical features of spina bifida neuropathic bowel and bladder dysfunction arguably have the greatest impact on social integration. The manifestations of bowel dysfunction in these children generally involve altered sensation of the presence of stool or fullness in the anal vault and/or decreased to absent external anal sphincter contractility.[2] Traditional management of these defecation problems is based on dietary modification supplemented by stool softeners or cathartics. While this approach may be successful in some children, documentation of its efficacy is lacking, perhaps reflecting the variable response to a problem that exists on a continuum of constipation to diarrhea. To this end other modalities have been used in an attempt to impact positively on neurogenic bowel dysfunction, namely biofeedback techniques, which have also demonstrated variable success,[3-5] and more invasive approaches, including enema continence catheters[6-8] and neuroprostheses.[9]

We previously demonstrated the efficacy of transurethral bladder electrostimulation on neurogenic bladder dysfunction in the myelomeningocele population, specifically improved bladder capacity and sensation.[10-12] The dependence of urination and defecation on neural pathways from spinal segments S2 to 4 led us to investigate the possible therapeutic efficacy of transrectal bowel electrostimulation on defecation in this population of children. We report encouraging results of this less invasive and more efficacious treatment of neuropathic bowel dysfunction in children with myelomeningocele.

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MATERIALS AND METHODS

A prospective study approved by the institutional review board at our hospital was performed on children with stool incontinence and myelomeningocele of various low spinal cord levels that were closed at birth. In all children suppositories or enema protocols were discontinued but the usual diets were maintained. Children were excluded from study if they had enteritis or a history of fistulas, or bowel dysfunction was successfully managed by another program. Candidates for electrical bowel stimulation therapy were fecally incontinent, not continent and on a failing bowel program.

Transrectal electrostimulation was performed using a stimulator with a 1.2 volt rechargeable battery. Sessions were done on an outpatient basis in the urodynamics suite under the supervision of nurses trained by 2 of us (I. R. and W. E. K.). The child was acclimated to the urodynamics suite before the session. The transrectal probe was then inserted 2 to 3 cm. into the rectum and a 5-second pulse of current (15 to 20 Hz. frequency and 2 to 3 mAmp. intensity) was delivered at 3 to 4-second intervals. Each session was performed for 30 minutes daily and 5 days weekly for 2 to 3 weeks. If benefits in any described parameter were noted, another series of 5 to 10 daily sessions was performed. If no benefits were noted after the first series of electrostimulation, therapy was discontinued.

The principal goal of electrostimulation is to provide bowel function in children with myelodysplasia that approaches

that of children without neurogenic bowel dysfunction. Electrostimulation was deemed completely successful when improvement was noted in all parameters of interest, while moderate success included improvement in any but not all parameters. Treatment failure was defined as a lack of improvement in any parameter. There were 4 parameters evaluated. 1) A decrease in the number of stooling episodes daily was defined as a reduction of at least 50% from a pre-therapy minimum of 6 stools daily in children older than 3 years and a reduction to 1 bowel movement daily in children younger than 3 years. 2) Increased sensation for having a bowel movement was noted when a child who previously did not have this sensation was able to discern this need intermittently and a child who previously discerned the need occasionally was able to do so regularly. 3) The ability to hold consciously a bowel movement was noticed at home by the parent or child as it related to improvement in the frequency of soiled underwear or accidents. 4) Subjective assessment was made by parents and/or children that there were significant changes in bowel habits since the start of electrostimulation based on pre-therapy expectations. Children and parents were surveyed on these parameters as well as any untoward effect noted during the electrostimulation sessions or at home following treatment. Statistical analysis was performed using a 1-sample test for binomial population, assuming a 50% chance of success, and with $p < 0.05$ considered significant.

RESULTS

A total of 55 children 2 to 14 years old (mean age 6.7) satisfied the criteria for study participation and they are included in this report. Each child was fecally incontinent with 6 to 15 daily bowel movements and not socially continent of stool. Gender distribution was nearly equal (28 boys and 27 girls). Children completed between 3 and 39 daily sessions (mean 18 sessions per patient) for a total of 996 sessions. Only 8 patients received fewer than 5 therapy sessions. There were no untoward effects during any treatments and none reported by parents at followup interviews. Followup ranged from 1 to 6 years.

Based on the established success criteria, transrectal electrostimulation was completely successful in 20 children (36.3%) and moderately successful in an additional 30 (54.5%, overall success rate 90.8%). Five children (9.1%) had no improvement in any parameter of bowel dysfunction after electrostimulation and treatment was considered to have failed (fig. 1). Two of these children underwent fewer than 5 daily sessions because of logistic problems with arriving for therapy. Patients who had success with electrical bowel stimulation did not require cathartics, suppositories or enemas after therapy and those in whom electrostimulation failed returned to the bowel program used before participation in our program.

Figure 2 shows the effects of electrostimulation on each parameter of interest. Specifically, 71% of patients reported complete bowel continence, while an additional 16% reported a decrease of at least 50% in the number of daily stooling accidents. Increased sensation for having a bowel movement was reported by 64% of the patients, while an additional 18% had improved sensation to a lesser degree. Improvement in the ability to hold a bowel movement was reported by 71% of the patients. Overall 53% of the children reported significant improvement in bowel function since the start of electrostimulation therapy and an additional 15% noted some improvement. The success (any degree of improvement) of transrectal electrostimulation was statistically significant for all 4 parameters ($p < 0.05$) and it was significant for success as defined for increased sensation ($p = 0.01$), ability to hold ($p = 0.02$) and bowel continence ($p = 0.0008$, see table).

DISCUSSION

Neurogenic bowel dysfunction is a major obstacle to social integration of the child with myelomeningocele. Management of these problems in defecation, ranging from chronic constipation to diarrhea, typically comprises nutritional and pharmacological modulation. Because the success of this management approach varies, other modalities have been used in an attempt to improve defecation. Biofeedback has been used to assist in improving sensation and sphincter control but its reported success is predicated on the active cooperation and motivation of the patient.[3,5] In a controlled study of a small population by Loening-Baucke et al the number of stooling episodes improved after 12 months but no patient learned to contract the external sphincter after biofeedback.[4] The search for other treatment modalities, such as selective sacral electrostimulation, has led to more invasive therapies designed to improve the outcome of bowel dysfunction but results are insufficiently positive to advocate routine application.[6-9]

The physiological importance of the sacral spinal cord to bowel and bladder function, particularly S2 to 4 from which the pudendal nerve originates, is well established from studies of selective sacral nerve root electrostimulation.[13,14] How-

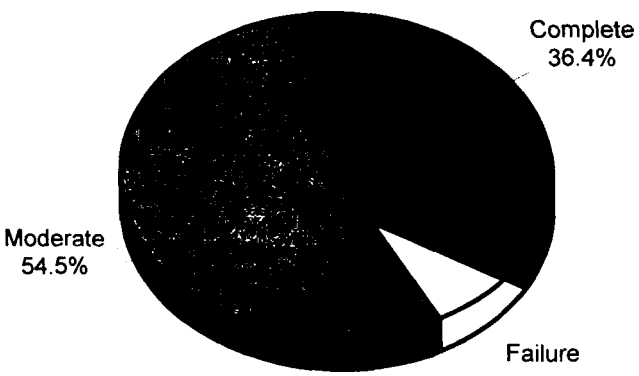

FIG. 1. Success rates of transrectal electrostimulation therapy in children with myelomeningocele and diarrhea. Overall success rate was approximately 91%. *Complete*, complete success. *Moderate*, moderate success.

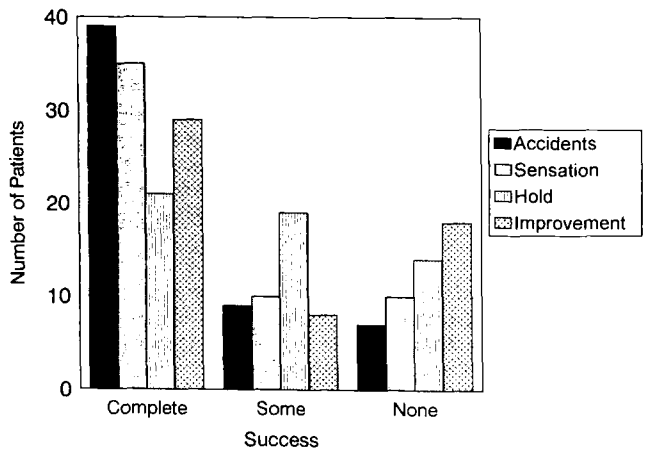

FIG. 2. Histogram of outcomes of 55 children undergoing transrectal electrostimulation therapy with respect to 4 parameters of interest. Decrease in number of daily bowel movements (*Accidents*) was most improved parameter, while subjective assessment of significant improvement in overall bowel function was least improved but greater than 80%. *Complete*, complete success. *None*, treatment failure.

One-sample test for binomial population p values, assuming a 50% chance of success

	Complete Success	Moderate Success
Fewer accidents	0.0008	<0.0001
Increased sensation	0.0140	<0.0001
Ability to hold	0.0234	0.0019
Observed improvement	0.1061	0.014

ever, clinical manifestations of these lesions vary and they depend on the degree of abnormal function of the pelvic floor musculature, external anal sphincter, perianal skin and anorectal mucosal sensation.[2] While the invasive nature of implantable neuroprostheses and difficulties inherent in operating on patients with myelomeningocele makes this option less attractive for managing neuropathic bowel dysfunction, electrical stimulation has been shown to be efficacious for improving bladder control, ejaculation and striated muscle spasticity in patients with spinal cord injuries or diseases. We previously reported the efficacy of transurethral bladder stimulation for increasing bladder capacity and improving the sensation of bladder fullness for bladder dysfunction in the myelomeningocele population.[10-12] These promising results with the understanding of the common neural pathway for bowel and bladder function led us to investigate a possible extension of electrostimulation therapy.

Ideally the paramount therapeutic goal in these children is to attain bowel function similar to that of children without neuropathic bowel dysfunction. We recognize that at best social control of neuropathic bowel dysfunction may be achieved by decreasing the number of daily bowel movements, increasing the ability to hold stool within the rectal vault and improving sensation independently or in various combinations. The best reflection of having achieved these goals is the maintenance of no need for diapers or pads because of stool problems. A total of 14 children of at least toilet training age (older than 3 years) is now free of such protective devices. Of the remaining patients 91% had improvement in at least 1 parameter of bowel dysfunction and 36% had improvement in all 4 parameters. Complete bowel continence or the elimination of stooling accidents was most often improved (71% of cases), followed by increased sensation (64%), which is a significantly better outcome than reported with biofeedback. Statistically the probability that these positive results were due to chance is less than 1% ($p < 0.01$). The success rate of this therapy may be underreported, since of the 8 children who underwent only 5 or fewer sessions 4 had complete success, 2 had moderate success (improved sensation in 1, and fewer bowel movements and improved ability to hold stool in 1) and treatment failed in 2. Transrectal electrostimulation was performed on an outpatient basis, and it was well tolerated and associated with no untoward effects. Moreover, at 1 to 6 years of followup all families reported maintenance of the positive effects of therapy. No child required repeat electrostimulation therapy after completing the course.

Another factor that impacts on the success rates is the preparedness and motivation of the child to move out of diapers. There were patients in whom stooling frequency decreased to 1 from 6 daily bowel movements but who remained in diapers because they were not prepared emotionally/developmentally to become toilet trained. These children may have complete success later without additional electrostimulation therapy. The youngest children (less than 2 years old) in this series had anal sphincter dysfunction to an extent that the sphincter was visibly open at physical examination. The goal of electrostimulation in these children was to decrease the number of daily bowel movements and improve sphincter tone with the prospect of improving or attaining continence as the child ages.

Success rates of electrical bowel stimulation were even better when only the more objective parameters were considered. The inclusion of parental subjective opinion decreased the success rate of this therapy but its inclusion is important to the overall evaluation of any modality. Parents asked whether this therapy improved bowel function in their child. One must recognize the high expectations that a parent may have for a new modality, particularly when it may impact on the most incapacitating social problem associated with myelodysplasia, and the potential disappointment when therapy efficacy fails to meet these high expectations. Future studies will concentrate on correlating the objective parameters of decreased bowel movements, sensation and the ability to hold stool with physiological parameters of biothesiometry and manometry.

Nevertheless, our results are encouraging for the management of neurogenic bowel dysfunction in children with myelodysplasia. In many children conservative measures succeed in managing bowel dysfunction, namely cathartics, bulking agents, enemas and digital stimulation. However, the cases in our study were limited to those in which more conservative approaches to bowel dysfunction were failing, and in this regard these more recalcitrant cases serve as controls. A prospective blinded study would provide unequivocal proof of the efficacy of electrostimulation therapy. However, the logistical difficulties associated with recruiting patients to such a study, given the prospect of randomization to the placebo arm (placement of the probe without transmission of electrical current) would be difficult, considering the minimal 2 to 3-week required time.

Few reports of the clinical use of bowel electrostimulation have been published and to our knowledge none exclusively describes children or the myelomeningocele population. Pescatori et al reported positive results in 10 of 15 patients with partial fecal incontinence, manifested by improved voluntary contraction of the external sphincter and emotional acceptance of the procedure, but no change in sensation was noted.[15] In contrast, Scheuer et al observed no improvement of internal or external sphincter function following electrostimulation.[16] While our study did not include manometric evaluation, improvement in sensation and the ability to hold stool as well as the elimination of stooling accidents reflects improvement in sensation and/or sphincter control. Ability to hold stool was the least commonly improved parameter (39% of cases) but it was reported to improve at least somewhat in 74%, perhaps reflecting a difference in the neuropathology of the myelomeningocele patient and the patient with pelvic floor denervation. In conclusion, transrectal electrostimulation is a well tolerated, minimally invasive modality that provides sustainable improvement in stool continence in patients with myelomeningocele and neuropathic bowel dysfunction to the point of eliminating the need for diapers or pads.

REFERENCES

1. Williamson, G. G.: Children with Spina Bifida: Early Intervention and Preschool Programming. Baltimore: Paul H. Brookes Publishing Co., 1987.
2. Younoszai, M. K.: Stooling problems in patients with myelomeningocele. South. Med. J., **85:** 718, 1992.
3. Wald, A.: Use of biofeedback in treatment of fecal incontinence in patients with myelomeningocele. Pediatrics, **68:** 45, 1981.
4. Loening-Baucke, V., Desch, L. and Wolraich, M.: Biofeedback training for patients with myelomeningocele and fecal incontinence. Dev. Med. Child. Neurol., **30:** 781, 1988.
5. Gil-Vernet, J. M., Marhuenda, C., Sanchis, L. and Boix-Ochoa, J.: Biofeedback in spina bifida. Ped. Surg. Int., **7:** 30, 1992.
6. Shandling, R. A. and Gilmour, R. F.: The enema continence catheter in spina bifida: successful bowel management. New York: Grune and Stratton, Inc., p. 271, 1987.
7. Malone, P. S., Ransley, P. G. and Kiely, E. M.: Preliminary report: the antegrade continence enema. Lancet, **336:** 1217, 1990.

8. Liptak, G. S. and Revell, G. M.: Management of bowel dysfunction in children with spinal cord disease or injury by means of the enema continence catheter. J. Ped., **120:** 190, 1992.
9. Schmidt, R. A., Kogan, B. A. and Tanagho, E. A.: Neuroprosthesis in the management of incontinence in myelomeningocele patients. J. Urol., **143:** 779, 1990.
10. Cheng, E. Y., Richards, I., Balcom, A., Steinhardt, G., Diamond, M., Rich, M., Donovan, J. M., Carr, M. C., Reinberg, Y., Hurt, G., Chandra, M., Bauer, S. B. and Kaplan, W. E.: Bladder stimulation therapy increases bladder capacity: results from a multi-institutional trial. J. Urol., part 2, **156:** 761, 1996.
11. Kaplan, W. E.: Alternatives to enterocystoplasty: bladder stimulation. Prob. Urol., **8:** 410, 1994.
12. Kaplan, W. E., Richards, T. W. and Richards, I.: Intravesical transurethral electrostimulation to increase bladder capacity. J. Urol., part 2, **142:** 600, 1989.
13. Shafik, A.: Sacral root stimulation for controlled defecation. Eur. Surg. Res., **27:** 63, 1995.
14. Martinez-Piñeiro, L., Trigo-Rocha, F., Hsu, G. L., Lue, T. F., Schmidt, R. A. and Tanagho, E. A.: Response of bladder, urethral and intracavernous pressure to ventral lumbosacral root stimulation in Sprague-Dawley and Wistar rats. J. Urol., **148:** 925, 1992.
15. Pescatori, M., Pavesio, R., Anastasio, G. and Daini, S.: Transanal electrostimulation for fecal incontinence: clinical psychologic, and manometric prospective study. Dis. Colon Rectum, **34:** 540, 1991.
16. Scheuer, M., Kuijpers, M. C. and Bleijenberg, G.: Effect of electrostimulation on sphincter function in neurogenic fecal incontinence. Dis. Colon Rectum, **37:** 590, 1994.