Case Study: Pudendal Nerve Stimulation in a Pediatric Patient

Section of Urology

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N euromodulation of the sacral nerve, or sacral nerve stimulation, is still rare but increasingly used in the pediatric population to help control fecal and urinary incontinence when other medical management has failed. Nationwide Children's Hospital has used sacral nerve stimulation to treat more than 60 individual patients since 2012. A significant number of patients with refractory bowel/bladder dysfunction, however, will have underlying comorbidities.

Patients with caudal regression can have partial or total agenesis of the sacrum, making typical sacral neuromodulation impossible. Neuromodulation of the pudendal nerve may be an option for those patients. While there are a handful of studies reporting on pudendal neuromodulation in adults and one Belgian study discussing pediatric cases, we are unaware of the treatment being used on a child in this country previously.

Here we present the case of a 9-year-old girl we believe to be the first pediatric patient in the United States to be treated with pudendal neuromodulation.





Image A shows the typical sacral nerve stimulator lead placement via the standard approach through the S3 foramen.



Image B shows the ischiorectal approach necessary in a pudendal nerve stimulator lead placement. Note the partial sacral agenesis seen in this patient.

Presentation:

A 9-year-old girl with a history of anorectal malformation, caudal regression and refractory bowel/bladder dysfunction presented to the Center for Colorectal and Pelvic Reconstruction at Nationwide Children's as a possible candidate for sacral neuromodulation. She had previously undergone Malone antegrade continence enema, appendicovesicostomy and bladder outlet reconstruction. Despite anticholinergic therapy and daily enemas, the patient still had significant fecal incontinence and severe bladder spasms that required periodic Botox injection into the bladder.

Magnetic resonance imaging demonstrated dysgenesis of the sacrum below S2, meaning she was not a candidate for standard sacral neuromodulation, which requires lead placement into the S3 foramen. Previous electromyography (EMG) had confirmed pudendal nerve function. We researched pudendal stimulation as a treatment used for adults, and talked with the patient's family about the possibility of adapting that procedure for a child.

Treatment:

Pudendal nerve stimulation uses the same implantable impulse generator and tined electrode lead as sacral nerve stimulation, but the positioning of the lead is different. In this case, a nerve localization technique was performed to place the lead into Alcock's canal adjacent to the pudendal nerve. Stimulation was confirmed by anal wink and compound muscle action potential monitoring with intraoperative EMG.

After this first phase of the procedure, the patient was given an external impulse generator for a two-week trial of the stimulation. The patient's episodes of fecal incontinence decreased significantly and her bladder spasms resolved. The second phase was then completed by placing the implantable impulse generator under the skin of the upper buttock.

Follow-up and Outcome:

The patient tolerated the procedure well, and after an approximately three-week recovery period, was allowed to resume normal activity. Two months post-procedure, her follow-up urodynamic study displayed a mildly improved bladder capacity, good compliance and no detrusor overactivity. Her compound action potential response was evaluated with EMG and impulse generator settings were slightly adjusted. After these setting changes, her remaining fecal incontinence has essentially resolved.

The patient, who was having daily fecal accidents before the procedure, is now able to wear regular underwear and engage in sports six months post-procedure. Pudendal neuromodulation was an incredibly effective treatment for her when all other conventional measures had failed.

Learning the Procedure:

To learn how to place the lead and properly evaluate compound action potential response for the pudendal nerve, four members of the Nationwide Children's neuromodulation team traveled to Michigan to observe Kenneth M. Peters, MD, perform three of these procedures on adults. Dr. Peters, Chief of Urology at Beaumont Hospital in Royal Oak, pioneered the use of pudendal stimulation in adults with refractory bladder dysfunction as well as chronic pelvic pain.

Placing the lead in a pudendal neuromodulation case is much more technically challenging than in a sacral neuromodulation case, and even surgeons with significant sacral nerve stimulation experience may have difficulty without observing the procedure first. The first phase of a sacral stimulation procedure typically lasts 90 minutes; the first phase in this case lasted three hours. Intraoperatively, nerve stimulation and function must be confirmed with EMG.

Possible Candidates:

Pudendal neuromodulation should be reserved for very complicated patients who have failed traditional management and, due to their complex anatomy, are not candidates for sacral neuromodulation. Very few patients will meet these criteria, and most will have already undergone reconstructive surgery for their bowel/bladder dysfunction. It is not clear if children with complete sacral agenesis, spinal cord injury or myelomeningocele would benefit from this procedure since they may not have adequate pudendal nerve function.

Since the procedure described in this case study, we have treated one other patient with pudendal neuromodulation. This second patient had part of her sacrum removed during surgery as a young child and was experiencing bladder deterioration and difficulty emptying. In the weeks since her procedure, her bladder residuals have improved, but her longer-term outcome is not yet known.

Because pudendal neuromodulation is not a well-known treatment, identifying future candidates may be challenging. Education and communication among gastroenterologists, urologists, colorectal specialists and others will be important for advancement in this area.



Seth A. Alpert, MD, is a member of the Section of Pediatric Urology at Nationwide Children's Hospital and a Clinical Associate Professor of Urology at The Ohio State University College of Medicine. Dr. Alpert serves as the Co-Director of the Surgical Neuromodulation Center as well as the Associate Program Director of the Pediatric Urology Fellowship Program. He received his medical degree from George Washington University School of Medicine, Washington, D.C. He completed urology residency at the University of Tennessee, Memphis, followed by a two-year pediatric urology fellowship at Ann & Robert H. Lurie Children's Hospital of Chicago. Dr. Alpert is interested in all

aspects of urologic problems in children including such diverse conditions as urinary tract infections, hydronephrosis, urinary incontinence, hypospadias, kidney stones and ureteral reflux.

Dr. Alpert also is a member of the Center for Colorectal and Pelvic Reconstruction, Nephrology and Urology Research Affinity Group, Prader-Willi Syndrome Clinic and Spina Bifida Program at Nationwide Children's.

Referrals and Consultations

The Section of Urology at Nationwide Children's Hospital accepts referrals from across the U.S. and internationally.

Online: NationwideChildrens.org Phone: (614) 722-6200 or (877) 722-6220 Fax: (614) 722-4000

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