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Prolonged spinal and sacral neurostimulation in children with pelvic organ dysfunction: preliminary analysis

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ABSTRACT

Objective. To evaluate the clinical efficacy of long-term spinal and sacral programmable neurostimulation for pelvic organ dysfunction in patients with myelodysplasia and chronic dysfunction of the bladder and rectum.

Material and methods. A retrospective study included 32 children aged 1—17 years (mean 10.7) with myelodysplasia, pelvic organ dysfunction and ineffective therapy including botulinum therapy and exclusion of tethered spinal cord syndrome. All children underwent comprehensive urodynamic examination with analysis of bladder and residual urine volume, mean flow rate, intravesical pressure and total urine volume, as well as electromyographic examination. Examination was carried out before surgery, after 6, 12 and 36 months. We applied urinary diary, NBSS questionnaire and urodynamic examination data. All patients underwent neurological examinations (neurological status, magnetic resonance imaging of the spinal cord, computed tomography and radiography of the spine, electroneuromyography). The study was conducted at the neurosurgical department of the Republican Children's Clinical Hospital in Ufa between 2014 and 2022. There were 32 implantations of epidural neurostimulators for pelvic organ dysfunctions.

Results. *P*atients used epidural spinal and sacral stimulation up to 6 times a day for 10—15 min turning on the pulse generator. This method significantly increased urinary volume, decreased episodes of urinary leakage and fecal incontinence, residual volume after urination and number of periodic catheterizations compared to baseline data. Sixteen patients were very satisfied, 10 ones were moderately satisfied, and 2 patients were not satisfied with therapy. The number of bladder catheterizations per day decreased by 51.1%. Urine volume significantly increased from 131.5±16.1 to 236±16.7 ml, intravesical pressure decreased from 23.5±4.2 to 18.5±2.1 cm H_2O (by 20.3%).

Conclusion. Chronic epidural spinal and sacral stimulation can improve the quality of life in patients with pelvic organ dysfunction. This technique may be effective for pelvic organ dysfunction caused by myelodysplasia.

Keywords: *sacral neuromodulation, spinal neuromodulation, pelvic organ dysfunction, children.*

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Abbreviations

SCI — spinal cord injury
ENMG — electroneuromyography

Introduction

Neurogenic dysfunction of the bladder and rectum is a complex and unresolved problem [1, 2].

Spinal cord and sacral root stimulation is one of the modern methods for chronic pelvic organ dysfunctions (bladder and intestines) [3].

In pediatric practice, congenital myelodysplasia is the most common cause of pelvic organ dysfunction (1 per 1000 newborns) [4, 5]. Acquired disorders occur in spinal cord injury (SCI), myelitis, and neurodegenerative diseases [5]. They lead to bladder dysfunction and, as a consequence, urinary tract infections, vesicoureteral reflux and renal failure [6]. According to the guidelines of the European Association of Urology, the concept of «neurogenic bladder» includes disorders of reservoir and evacuation functions of the bladder in combination with detrusor-sphincter dyssynergia following myelodysplasia [7]. A typical feature of neurogenic bladder in children is its combination with dysfunction of the colon and rectum (40—45% of patients) [8].

Modern methods for pelvic organ dysfunctions are often aimed only at eliminating symptoms and do not always improve the quality of life [3, 4]. Spinal and sacral stimulation is sometimes used in rehabilitation measures for these patients [9].

Effectiveness of neuromodulation for pelvic disorders was demonstrated back in the 80s of the last century, although appropriate mechanism was unknown [10]. Currently, carious studies confirm the effectiveness of neurostimulation in adults [3, 11, 12], and only a few studies emphasize the feasibility of this technique for neurogenic pelvic organ dysfunctions in children [4, 13, 14].

The purpose of the study was to evaluate the clinical efficacy of long-term spinal and sacral programmable neurostimulation for pelvic organ dysfunction in patients with myelodysplasia and chronic dysfunction of the bladder and rectum.

Material and methods

A retrospective study included 32 patients with myelodysplasia and secondary neurogenic pelvic organ dysfunctions. All patients underwent implantation of systems for chronic spinal and sacral stimulation in the neurosurgical department of the Republican Children's Clinical Hospital of the Republic of Bashkortostan between March 2014 and August 2022. The local ethics committee approved the study. All patients signed an informed consent for intervention.

We enrolled patients with myelodysplasia and drug-resistant pelvic disorders undergoing surgery for meningoradiculomyelocele, lipomeningocele and tethered spinal cord. There were 16 girls and 18 boys. Mean age was 11.2±4.1 years. Exclusion criteria were urinary tract obstruction, stress-induced incontinence and mental disorders.

Indications for chronic neurostimulation were chronic non-obstructive retention or urge urinary incontinence, failure of conservative treatment. There were 32 appropriate patients including 27 ones with meningoradiculomyelocele and lipomeningocele, 5 ones after correction of tethered spinal cord. These patients underwent previous excision of meningoradiculomyelocele, lipomeningocele with hernia repair and correction of tethered spinal cord. All patients completed voiding diaries and underwent urodynamic and neurological examinations.

Examination protocol. Patients underwent urological and neurological examinations before neurosurgical treatment (consultation with a proctologist if necessary). We analyzed neurological status, MRI of the spinal cord, CT and X-ray examination of the spine, electroneuromyography (ENMG) data.

The main functional diagnostic method was urodynamic testing including bladder volume and bladder pressure at maximum filling, volumes of urine excreted and residual urine. In addition, a voiding diary was kept. Examination was carried out before surgery, after 6, 12 and 36 months.

Finally, we distinguished 2 groups of patients: detrusor hypotonia (20 patients) and detrusor hyperactivity (12 patients).

Total urinary incontinence, lack of urge to urinate, anal sphincter insufficiency and stool, constipation prevailed in the 1st group. Urodynamic testing revealed insufficiency and decreased tone of urethral and anal sphincters, large amount of residual urine and electromyography data on severe peripheral conduction disorders. In the 2nd group, we observed daily episodes of urinary incontinence (paradoxical ischuria), frequent and painful urge to urinate, inability to «hold» urine, stool incontinence and constipation. Urodynamic testing revealed decrease of bladder volume, increased intravesical pressure and detrusor-sphincter dyssynergy. There were ENMG data on axonal disorders.

We used epidural neurostimulators Prime ADVANCED, Prime Advanced Sure Scan MRI with Vectors 1×8, Stim wave Freedom, INTERSTIM II electrodes for pelvic organ dysfunction. The last ones were implanted at the lower thoracic and upper lumbar levels (n=16) or in sacral region (n=16).

After determining the indications for neurosurgical treatment, we installed a permanent system for neurostimulation. The test period of neurostimulation (1—2 weeks) in children with neurogenic pelvic organ dysfunction is insufficient. In our opinion, a longer period (up to 6 months) is needed to select neurostimulation parameters for a particular patient and assess effectiveness of this technique.

The level of electrode depended on anatomical features (anomalies of the sacrum and spine). Thus, we chose the lower thoracic and upper lumbar levels if insertion of sacral electrode was technically impossible (sacral agenesis, scars). Surgery was performed under general anesthesia in an operating room equipped with fluoroscopic devices **(fig. 1, 2)**. We confirmed correct installation of electrode considering contractions of the anal sphincter and dorsiflexion of the big toe.

After procedure, patients were programmed with their device and trained to use the programmer. Patients completed a voiding diary and short satisfaction questionnaire at the follow-up visit. We compared the last completed voiding diary with baseline one. Control urodynamic testing were performed in 6, 12, 36 months after surgery.

The criteria for selecting neurostimulation parameters were feeling of bladder filling, normalization of rhythm and frequency of urination. Mean neurostimulation parameters in children: frequency 15—55 Hz, pulse duration 80—200 ms, strength — 0.5—4.4 V.

After discharge, children are followed-up by neurosurgeon, neurologist, urologist, and surgeon. They undergo periodic urological examinations (ultrasonography, urodynamic testing) at least once every 6 months. Correction of electrical stimulation parameters is required within the first 2—3 months due to local tissue reaction and fi-

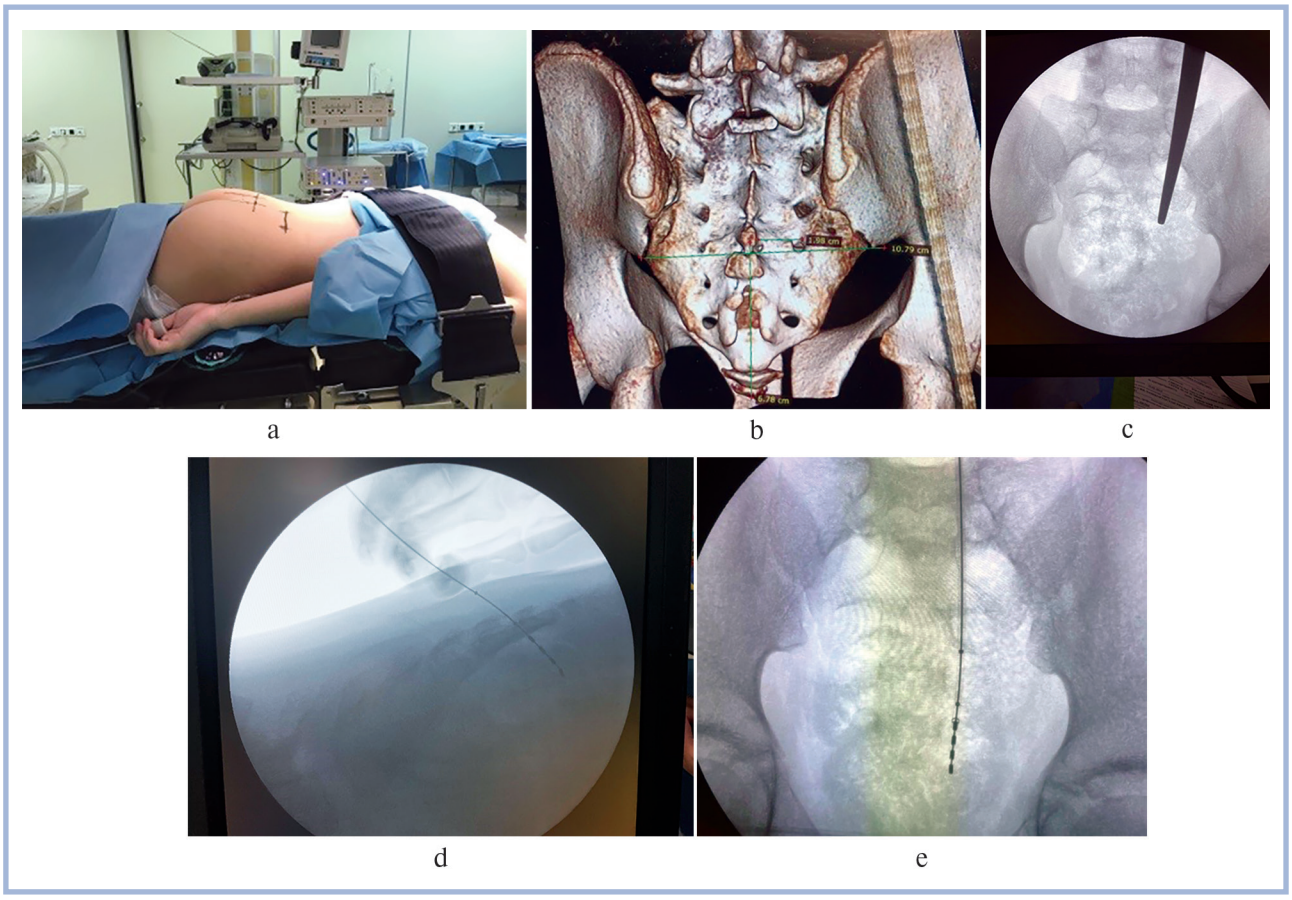

Fig. 1. **Sacral neuromodulation procedure.**
a — positioning the patient and surgical field marking; b — intraoperative radiography of the sacrum; c—d — implantation of sacral electrode under X-ray control.

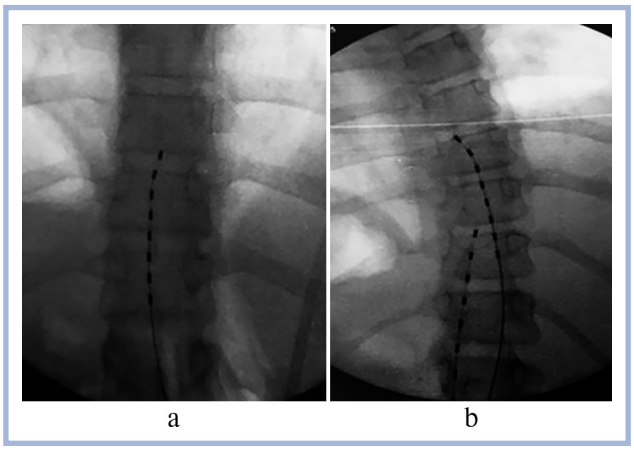

Fig. 2. **Spinal stimulation.**
a, b — X-ray pattern of the implanted spinal electrodes.

brosis around the electrode. Quality of life was assessed using the Neurogenic Bladder Symptom Score (NBSS) questionnaire [2, 15].

We did not calculate the sample size. We also recognize that small number of samples can affect statistical significance of results. Data were tested for normal distribution using SPSS histogram. We compared continuous variables by using of the Mann—Whitney *U* test, qualitative variables — χ^2 values. Two-sided *p*-value <0.05 was significant.

Results

Treatment outcomes were assessed throughout 6—84 months after implantation **(table 1—3)**.

Constant spinal and sacral neuromodulation therapy led to subjective improvement of the quality of life in almost all patients (according to urination diary). There were significantly more common urges to urinate and no need for bladder catheterization. Most patients normalized defecation. Bladder volume significantly ($p<0.05$) increased, while the volume of residual urine became lower. The NBSS score decreased significantly ($p<0.01$).

When comparing permanent spinal and sacral neurostimulation therapy, we found no significant differences in effects of permanent epidural neurostimulation depending on the level of stimulator.

There was a significant ($p<0.05$) decrease in mean urine flow rate and increase in bladder volume before surgery in children with hypoactive bladder. Urodynamic testing revealed no significant between-group differences after chronic neurostimulation. In all patients, all uro-

Table 1. **Treatment outcomes after neurostimulation**

Criterion	Thoracic $Th_{X–XII}$, lumbar $L_{I–II}$ electrode (*n*=16)		Sacral S_{III} electrode (*n*=16)		Total	
	n	%	*n*	%	*n*	%
Feeling the urge to urinate	10	62.5	15	93.8	25	78.1
No need for bladder catheterization	7	43.8	12	75.0	19	59.4
Feeling of bladder fullness	10	62.5	8	50.0	18	56.2
Improvement of urodynamic parameters (bladder volume, residual urine, mean urine flow rate, urine output, intravesical pressure at maximum filling)	15	93.8	15	93.8	30	93.8
Subjective improving the quality of life	15	93.8	16	100	31	96.8
NBSS score	9.4±4.2		9.1±3.9		9.2±4.1	
Defecation	10	62.5	9	57.1	19	59.4
Concomitant positive effects						
Healing of trophic foot ulcer	2	—	0	—	2	—
Lower sweating of the lower limb, feeling of comfort in the lower limb	12	—	6	—	18	—

Table 2. **Urodynamic testing in patients with hypoactive bladder**

Urodynamic testing data	Before surgery (*n*=20)	After surgery (*n*=20)
Bladder volume, ml	66±12	158±15*
Residual volume, ml	80±20	10±4.9*
Mean urine flow rate, ml/s	3.8±1.2	16.5±4.1*
Volume of urine excreted, ml	143±15	242±15*
Intravesical pressure at maximum filling, cm H_2O	23±5	18.0±5

Note. Here and in **table 3**: * — $p<0.05$.

Table 3. **Urodynamic testing in patients with hyperactive bladder**

Urodynamic testing data	Before surgery (*n*=12)	After surgery (*n*=12)
Bladder volume, ml	40.1±19.2	182±10.1*
Residual volume, ml	30.0±10.1	10±5.2*
Mean urine flow rate, ml/s	13.2±4.0	18.2±3.1
Volume of urine excreted, ml	120±25.4	230±25.7*
Intravesical pressure at maximum filling, cm H_2O	34.2±10.0	19.0±10.2*

dynamic parameters significantly improved **(table 2, 3)**, with the exception of mean urine flow rate in children with overactive bladder.

There were 2 complications, i.e. bedsore within the generator area and epidural electrode migration. Complications were eliminated in all cases.

To confirm our results, we present 2 clinical examples.

Clinical example No. 1

A 14-year-old patient M. underwent surgery for lumbosacral meningoradiculocele at the age of 1 month in 2018. Fecal and urinary incontinence persisted after surgery. The child was treated in various hospitals for urological problems; repair of ureteric orifices and urethral stenting were performed. ENMG revealed conduction disorders along *n*. tibialis dex/sin and *n*. peroneus dex, as well as decreased amplitude of proximal M-responses. Urodynamic testing revealed hypoactive bladder **(table 4)**. The Prime ADVANCED system with 8 contact electrodes was implanted at the ThXI—XII level on August 3, 2018. Urodynamic testing after 6 months **(table 4)** revealed no fecal and urinary incontinence without catheterization of the bladder.

Table 4. **Urodynamic testing data**

Variable	Before surgery	After surgery
Mean urine flow rate, ml/s	3.2	6.5
Volume of urine excreted, ml	80.2	182
Residual urine volume, ml	80	0
Maximum bladder volume, ml	143	242
Intravesical pressure at maximum filling, cm H_2O	23	18.0

Clinical example No. 2

A 13-year-old patient was diagnosed with myelodysplasia, anomaly of the sacrum, agenesis of the coccyx, tethered filum terminale and lipoma of the filum terminale. There were previous surgical interventions for pelvic organ dysfunction and neurogenic overactive bladder, fecal incontinence. The Interstim II system for sacral stimulation was installed in May 2021. There was obvious clinical effect by August 2022 (feeling of fullness of the bladder, reduced volume of residual urine, no episodes of urinary infection over the year) **(table 5)**.

Table 5. **Urodynamic testing data**

Variable	Before surgery	After surgery
Mean urine flow rate, ml/s	13.2	8.2
Volume of urine excreted, ml	60.2	182
Residual urine volume, ml	52	10
Maximum bladder volume, ml	120	230
Intravesical pressure at maximum filling, cm H_2O	34.2	18.0

Discussion

This study demonstrated a certain effectiveness of chronic neurostimulation in rehabilitation of children with congenital myelodysplasia accompanied by neurogenic pelvic disorders. Success rate of this procedure was 71.9% (23 out of 32 patients). The results were better in patients with overactive bladder (83.3%; 10 out of 12 patients) compared to atonic bladder (65.0%; 13 out of 20 patients). This is probably due to better baseline quality of the nerves with subsequent better restoration and modulation of lost functions in postoperative period. Pelvic neurostimulation improves neuromuscular conductivity of perineal striated muscles due to neurotrophic effect. Thus, we confirm the data of foreign researchers on successful operations depending on the type of neurogenic dysfunction of the lower urinary tract [9, 10].

The quality of life improved in 96.8% of patients. It was slightly higher when the electrode was located at the sacral level. This may be due to the most anatomically and physiologically accurate SIII stimulation.

We observed intestinal sphincter recovery (no fecal incontinence and reduced duration of constipation) in 12 children. Other authors obtained similar data [11, 14, 16].

Our study had several limitations (small sample size, heterogeneous patient population and not all neurological diseases followed by pelvic disorders). Timing of assessment of clinical effectiveness of epidural neurostimulation was heterogeneous too. There was high patient satisfaction with treatment. Nevertheless, a prospective study of large sample is necessary to confirm clinical effectiveness of this technique.

Conclusion

1. Chronic epidural stimulation for pelvic dysfunction is an effective method of symptomatic treatment of neurogenic dysfunction of urination and defecation when other treatment methods are ineffective and in a complex of rehabilitation measures.
2. Pelvic functions are improved in lower thoracic-upper lumbar and sacral epidural neurostimulation.
3. Chronic sacral electrical stimulation may be effective for overactive bladder and hypoactive detrusor.
4. Chronic epidural spinal cord stimulation (lower thoracic and upper lumbar levels) may be effective for pelvic organ dysfunction if sacral implantation is technically impossible (sacral agenesis, scars).

Author contribution:
Concept and design of the study — Timershin A.G.
Collection and analysis of data — Kreshchenok D.V., Konovalov S.A.
Statistical analysis — Timershin A.G.
Writing the text — Timershin A.G.
Editing — Mironov P.I.

No conflict of interests to declare.

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COMMENT

The manuscript is devoted to chronic spinal and sacral neurostimulation for pelvic organ dysfunction in children with myelodysplasia. The abstract clearly states the objectives of the study. Preoperative and follow-up examinations are described in detail. The authors present urodynamic testing data. Literature review is devoted to neurogenic pelvic organ dysfunction in children and results of chronic sacral neurostimulation. The authors correctly note that most studies provide results of this procedure in adults. Then, the researchers describe 32 patients divided into 2 groups depending on neurogenic dysfunction (hypoactive and hyperactive detrusor). When describing the technique, the authors presented two options for implanting the electrodes (SIII root and lower thoracic level). The results were assessed over a period of 6-84 months.

There were no significant differences in urodynamic parameters depending on the level of electrode implantation. However, treatment outcomes were better in patients with detrusor overactivity. Other authors confirmed this fact.

We have a retrospective non-randomized study. Statistical analysis of differences implied the Mann—Whitney test. There are still a few studies in the world literature devoted to long-term results of chronic sacral neurostimulation for neurogenic pelvic organ dysfunction in children. Prospective placebo-controlled studies with long-term follow-up period are advisable. Importantly, the authors did not use a test period of neurostimulation. After establishing the indications for neurosurgical treatment, a permanent neurostimulation system was immediately installed. This seems to us not entirely correct, since electrical stimulation through a test electrode for 1—2 weeks allows us to assess effectiveness of this technique in a particular patient. This makes it possible to exclude patients resistant to chronic stimulation and improve long-term results.

The authors used two levels for electrode implantation (SIII root and lumbar enlargement of the spinal cord). The second option was used in case of technical difficulties for implantation through the sacrum (sacral agenesis, adhesions). In our opinion, it is advisable to use only one level of implantation for better statistical quality of results. If the objectives implied comparison of chronic sacral and spinal stimulation, two homogeneous groups of patients with similar age, disorders and sample size were necessary.

All these comments do not reduce practical significance of this study. Chronic stimulation in children with neurogenic pelvic organ dysfunction can improve the quality of life and avoid such a serious complication as chronic renal failure. The article is of practical interest for neurosurgeons, neurologists and urologists.

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