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Rehabilitation Protocols for Children with Dysfunctional Voiding

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Abstract

Dysfunctional voiding is a functional voiding disorder characterized by an intermittent uroflow rate due to involuntary intermittent contractions of the striated muscle of the external urethral sphincter or pelvic floor muscles (PFMs) during voiding in neurologically normal children. Symptoms include voiding difficulties as well as urgency, voiding frequency and, in some instances, urinary incontinence and/or nocturnal enuresis. Recurrent urinary tract infections, chronic constipation and/or fecal incontinence and vesicoureteral reflux (VUR) contribute to this condition. Urotherapy is the mainstay of the treatment. It starts with education and demystification and simple behavioral modifications. Specific measures include PFM exercises with various forms of biofeedback concentrating at the recognition of PFM function and their relaxation. However, the PFMs are part of the abdominal capsule and they act in coordination with lower abdominal muscles. These muscles need to be relaxed during voiding. Diaphragmatic breathing exercises were introduced to teach children abdominal muscle relaxation. Easy to learn exercises do not require any specific equipment and can be performed at all health care levels. Children from five years of age could benefit from these exercises. In children resistant to standard treatment, botulinum toxin type A application, intermittent catheterization and surgery in children with VUR are recommended.

Keywords: dysfunctional voiding, urotherapy, pelvic floor exercises, diaphragmatic breathing exercises, pelvic floor electromyography biofeedback

1. Introduction

Disorders of bladder and bowel control are among the most common problems in childhood. At the age of 7, 10% of children get wet during the night, 2 to 3% wet their clothes during the day, while 1 to 3% of children have fecal incontinence [1]. These disorders often occur together. Despite a high rate of spontaneous remission, 1 to 2% of adolescents have nocturnal enuresis and less than 1% have daily urinary incontinence or fecal incontinence [2]. Most of these disorders are functional, i.e. they are not caused by neurological, structural, or medical factors.

Functional voiding disorders are some of the main causes of daily wetting in children, the development of recurrent urinary tract infections and vesicoureteral reflux (VUR). In addition to the risk of developing structural changes in the bladder

wall and upper urinary tract, voiding disorders, accompanied by urinary incontinence, can be a severe psychosocial problem. Children describe wetting at school as the third most stressful event in life after the death of a parent and loss of sight [3]. It is evident that urinary incontinence causes significant psychological morbidity, and treatment is crucial.

Functional voiding disorders can be treated in a number of ways, including pharmacological therapy, urotherapy, and surgical treatment in the most severe cases.

2. Dysfunctional voiding

2.1 Definition

Dysfunctional voiding (DV) refers only to the disorder of the bladder emptying phase, and is characterized by intermittent contraction of the external urethral sphincter and/or PFMs during the voiding phase of a micturition cycle [4]. A typical finding is interrupted or staccato uroflowmetry curve with increased electromyography (EMG) pelvic floor muscle activity during urination.

The more severe form is referred to as Hinman syndrome by the author who first described it [5]. Other terms previously used for DV are detrusor-sphincter discoordination, non-neurogenic neurogenic bladder and occult neurogenic bladder. In the United States, the term “dysfunctional voiding” has been used for all types of voiding disorders, even bladder filling phase disorders. According to the ICCS standardization of terminology from 2016, DV refers exclusively to the disorder of the voiding phase [4]. It is thought to be the result of excessive PFM activity in an attempt to prevent urination that occurs due to uninhibited detrusor contractions in the early stage of bladder filling.

2.2 Epidemiology

Epidemiological data on DV are lacking. Dysfunctional voiding was found to occur in 4.2% of children referred for urinary incontinence [6]. In published studies, the prevalence was estimated to be between 5 and 25% and 32% [7, 8]. Dysfunctional voiding was observed in 65% of children aged 5-9 years with urinary tract infections, and in 23% of children who were urinary tract infections free [9]. It is evident that the criteria for including children in the studies were different, as well as the accuracy of their evaluation, which indicates the need to conduct new research to determine accurate data.

2.3 Etiology

Dysfunctional voiding was first observed in 1973 by Hinman and Baumann [5]. Hinman describes it as an acquired, reversible behavioral disorder that can be ameliorated by suggestion and changes in behavior. He defined it as a bad habit for special people in a bad family environment. Allen stated in 1977 that hyperactivity of a child is a typical sign and that psychological factors play a key role in at least 50% of the 21 children described [10]. He also points to the importance of stressful situations in the family, such as parental alcoholism, parental divorce and father dominance.

Contrary to these considerations, Van Gool points out that DV is not related to emotional or psychosocial problems, but is caused by delayed CNS maturation and external urethral sphincter dysfunction [11]. Hjalmas considers the importance of hereditary factors, as DV was observed in several members of the

same family [12]. It is not known whether this is due to genetic factors or common family habits.

Most authors, however, believe that DV is a learned behavior [13]. It can develop from overactive bladder (OAB) and voiding postponement as a result of PFM contractions in an attempt to prevent urination, but it can also exist without these precursors. In some children, urgency, voiding frequency and, in some instances, urinary incontinence and signs of DV are present at the same time [14].

From the review of the literature so far, it can be concluded that the etiology is probably multifactorial. Possible risk factors are:

1. Inadequate toilet training process

Wiener et al. suggest that functional voiding disorders may be caused by inadequate toilet habits and postures [15], which was also confirmed in the study by Bakker et al. [16]. This study indicated that the use of adult toilets in the process of children's toilet training may increase the risk of developing functional voiding disorders. Thus, most of the programmes used in the treatment of DV involve taking an adequate position when urinating, i.e. the use of a toilet insert and footrests to ensure the stability of the trunk and the relaxation of the PFMs, and thus enable the physiological emptying of the bladder.

2. Small structural anomalies in the anatomy or innervation of the lower urinary tract.

3. Delay in maturation: detrusor overactivity as a component of DV may represent the persistence of normal infantile mode of urination even after toilet training. It is possible that delays in CNS maturation reduce the ability of these children to take voluntary control of the micturition reflex.

4. Impact of school: more than half of the time children spend in school, which suggests that teachers can positively or negatively influence the acquisition of toilet habits. In a study by Cooper et al., the influence of schools on the development of DV in children was examined and it was pointed out that most teachers allow going to the toilet only during rest [17]. Such a ban on going to the toilet for a child with an urgency who has not yet developed a complete inhibition of the micturition reflex may impair the normal function of the urinary bladder and sphincter. In addition, most children with daily urinary incontinence avoided going to the school toilet due to lack of privacy or poor toilet hygiene [17].

5. Unpleasant events during toilet training and/or personal stress: serious emotional stresses, such as sexual abuse, mostly of girls, should be considered in the event of sudden onset of DV, and in the absence of other etiological factors [18].

2.4 Clinical signs and symptoms

Children are usually referred for wetting clothes, but not for DV. Children and parents usually do not register specific symptoms of DV, so the physician must insist on them. Typical signs are difficulty initiating micturition (hesitancy), as well as straining to overcome the resistance of the contracted urinary sphincter. The urinary stream is usually not strong because the PFMs do not relax completely, and it is often intermittent.

In an attempt to postpone voiding or suppress urgency and/or urinary incontinence, children assume characteristic positions such as crossing their legs, standing

on tiptoes, squatting, or manual compression of the genitals (pelvic holding maneuvers). It is typical for girls to squat by pressing their heel against the perineum [19].

Stool retention, chronic constipation and fecal incontinence occur in more than 50% of children as a result of repeated and habitual contractions of the PFMs [9].

Plenty of data from the literature indicate an association between DV and recurrent urinary tract infections [8, 20–22]. Treatment of DV reduces recurrent urinary tract infections. About 50% of children with DV may have VUR [10].

2.5 Hinman-Allen syndrome

The most severe form of DV is Hinman-Allen syndrome, according to the authors who first described it in 1973 [5, 10]. The old term “non-neurogenic neurogenic bladder” can also be found in the literature.

This syndrome occurs in children who voluntarily and habitually contract the external urethral sphincter during uninhibited detrusor contractions, which leads to the inability of emptying the bladder. The condition is characterized by detrusor overactivity and possibly its decompensation. It is clinically manifested by daytime and nighttime wetting, urgency and overflow incontinence, chronic constipation, as well as recurrent urinary tract infections. Voiding cystourethrography reveals a trabeculated bladder, high-grade bilateral VUR and large post-void residual urine. If not treated in time, it leads to reflux nephropathy followed by kidney scarring, hypertension and progressive renal failure.

3. Evaluation of a child with dysfunctional voiding

Evaluation of children with DV includes taking anamnestic data, physical examination, filling in the bladder and bowel diary, urinalyses and urine culture, ultrasound examination of the bladder and kidneys and uroflowmetry with determination of post-void residual urine. Cystometry, voiding cystourethrography and MRI of the spine are indicated only in certain cases.

3.1 History

The evaluation process begins with anamnesis, data on perinatal factors, developmental course, current mental state, school success and events during toilet training. Every child should be asked how he urinates, whether he has difficulty starting to urinate, whether urinating is difficult and whether he strains when urinating. Also, one should insist on the characteristics of the urinary stream, such as strength and continuity. A weak and intermittent stream indicates the existence of DV. The child should be asked if he has a feeling of incomplete urination or urinary retention (inability to urinate). Questions regarding urgency, voiding frequency and severity of daytime and nighttime wetting, pelvic holding maneuvers, and bowel emptying should follow.

3.2 Physical examination

3.2.1 Abdominal examination

Palpation of the left iliac fossa is necessary in order to determine fecal impaction. Suprapubic tenderness may indicate the presence of cystitis. If urine leaks when the bladder is pressed, the existence of a neurogenic bladder with sphincter damage should be suspected.

3.2.2 Neurological examination

It is necessary to perform a careful inspection of the lower part of the spine in order to determine the cutaneous manifestations of occult spinal dysraphism and/or sacral agenesis (lipomas, nevi, increased hairiness, low intergluteal cleft, flattened buttocks). After that, it is necessary to test the tendon reflexes on the lower extremities as well as the existence of the Babinski sign. Hyperreflexia, asymmetry in reflexes or positive Babinski indicate spinal cord damage. It is also important to examine the strength of the muscles of the lower extremities, ataxia and gait.

3.2.3 Inspection of genitals and perianal region

Genital inspection is necessary in every patient with DV. Inspection of the perineum reveals the position of the anus, fissure, fistula or perianal inflammation of the skin in children with constipation.

3.2.4 Rectal examination

Digital rectal examination can determine the tone and function of the anal sphincter, the width and content of the rectal ampulla, the amount and consistency of feces in the ampulla, and the presence of pain. It is recommended that anorectal examination be performed only in those children who meet 1 of 6 Rome III criteria for the diagnosis of constipation [9].

3.3 Bladder and bowel diary

The bladder diary is a crucial diagnostic and therapeutic instrument for children with voiding disorders. After the anamnesis and physical examination, this is the third most important part of the evaluation. It is used to understand and quantify the function of the bladder at home, because the memory of the elements of voiding is unreliable. A complete bladder diary is kept for 7 nights and urinary incontinence is registered, as well as nocturnal urine volume, in order to evaluate nocturnal enuresis [4].

The 48 hr. daytime frequency and volume chart is kept for 48 hours, preferably on weekends for practical reasons [4]. The data obtained are frequency of voiding, volume of individual urination, maximum voided volume, urine production, nocturia, nocturnal urine production and fluid intake. The frequency of voiding in children with DV is variable.

A bowel diary is kept for 7 days to rule out the presence of constipation [4]. The frequency of defecation, the appearance of the stool according to the Bristol scale, pain and tension during defecation, whether the child has previously delayed defecation and whether fecal incontinence is present are entered.

3.4 Urinalysis and urine culture

Urinalyses and urine culture are initial tests to rule out urinary tract infection.

3.5 Urodynamics

3.5.1 Uroflowmetry

Uroflowmetry is the simplest form of urodynamics. As a non-invasive method, it is ideal for pediatric needs (**Figure 1**). It enables the examination of the function of the detrusor and the sphincter in the voiding phase of the child's micturition.



Figure 1.
Uroflowmetry.

The uroflowmeter was first described in the 1950s [23]. The method can be applied from the fourth year of life of a child with established micturition control. In order to get reliable results, it is necessary for the child to urinate at a capacity that is not less than 50% nor more than 115% of the bladder capacity expected for age [4]. In addition, it is necessary to repeat examination to make the result accurate and reliable.

The method consists in urinating the patient in a uroflowmeter, continuously measuring the urine flow rate (ml/s) and at the same time graphically showing the curve. The placement of two superficial EMG electrodes on the perineum enables the recording of the activity of the PFMs in the micturition phase. In this way, significant data are obtained, especially in children with DV. The uroflowmetry curve is of the staccato or interrupted type with an increase in the EMG activity of the PFMs during urination (**Figure 2**).

Upon completion of this study, an ultrasound examination of the bladder is performed to determine post-void residual urine. In a child aged 7 to 12 years, residual urine greater than 20 ml or 15% of the bladder capacity expected for age is considered elevated [4]. Residual urine that is larger than 10 ml or 6% of bladder capacity expected for age during several measurements is also considered elevated [4].

3.5.2 Cystometry

Cystometry is the only method by which bladder function can be examined directly and in detail. The method is invasive. In order to obtain the data necessary

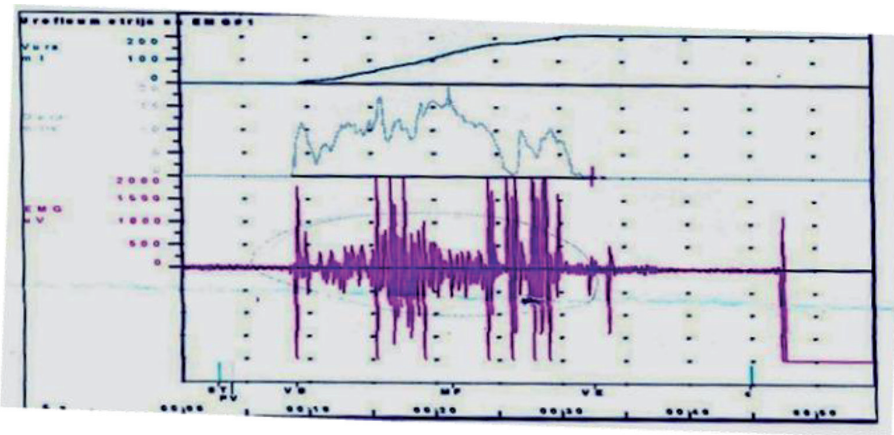


Figure 2.
Interrupted uroflowmetry curve with increased pelvic floor EMG activity during voiding.

for urodynamic analysis of bladder function, simultaneous measurement of pressure in the bladder, urethra and abdomen is required. This continuous measurement of detrusor pressure and sphincter activity during bladder filling and emptying allows an accurate diagnosis to be made for most lower urinary tract disorders.

In children with DV, cystometry is indicated only in certain cases, primarily because the diagnosis is made using non-invasive methods. It should be performed only in severe cases of DV, which should be differentially and diagnostically distinguished from neurogenic bladder with detrusor sphincter dyssynergia, suspicion of Hinman-Allen syndrome and DV resistant to the applied therapy.

3.6 Voiding cystourethrography (VCUG)

Voiding cystourethrography is not routinely performed in children with DV. It is used in children with recurrent urinary tract infections to rule out the presence of VUR. A typical finding in DV is a ballooned proximal urethra. If ultrasound examination reveals bladder wall thickening and hydronephrosis and/or a plateau uroflowmetry curve, it is necessary to perform VCUG to exclude the presence of a posterior urethral valve in boys.

3.7 Magnetic resonance imaging

Magnetic resonance imaging (MRI) of the spine should be performed in every child with a suspected neurogenic voiding disorder or in whom a clinical examination has established the existence of spinal dysraphism. Suspicion of tethered cord syndrome, as well as spinal tumors, are also indications for referring a child for an MRI of the spine.

4. Urotherapy

Functional voiding disorders should be actively treated due to the possibility of developing structural changes in the bladder and upper urinary tract. In the Anglo-Saxon literature, the term “urotherapy” has been used since 1980 in children with voiding disorders, which is synonymous with rehabilitation of the lower urinary tract [24].

4.1 Definition

Urotherapy is a conservative, non-surgical treatment of the lower urinary tract and can be defined as re-education of the bladder or rehabilitation programme

aimed at correcting the phase of filling and emptying the bladder [4]. Urotherapy, first of all, includes changing the habits that the child has acquired in the process of toilet training, as well as establishing motor control over the micturition reflex. It can also include drug therapy, because most children have some kind of pharmacotherapy during urotherapy.

4.2 Goals

The goals of urotherapy are clearly defined and include normalization of the phase of filling and emptying the bladder, facilitation of normal bowel function and normalization of the dynamics of defecation. The goals of treatment in clinical practice are aimed at reducing urgency and urinary incontinence, nocturnal enuresis, post-void residual urine, high intravesical pressure, PFM activity in the micturition phase, normalization of uroflowmetry curve, cure urinary tract infections and constipation, and reduce the degree of VUR.

4.3 Indications

Urotherapy is used in functional voiding disorders that are accompanied by an altered phase of bladder filling and emptying, in children in whom satisfactory results have not been achieved with the use of pharmacological therapy, and in children who are just starting therapy.

4.4 Components of urotherapy

Urotherapy consists of standard urotherapy and specific measures that include neuromodulation, PFM relaxation exercises, biofeedback and intermittent catheterization [4].

4.4.1 Standard urotherapy

In the literature, we often come across the term “standard urotherapy”, which includes education, behavioral modifications, keeping a bladder and bowel diary and regular check-ups [4]. Before starting urotherapy, it is necessary to dedicate a lot of time to educating the child, examining his motivation to carry out the treatment, and eliminating the shame and other effects that urinary incontinence can cause.

4.4.1.1 Education and demystification

The child should be acquainted with the structure and function of the bladder, external urinary sphincter, colon and anorectum in a way that is appropriate for his age. He also needs to be explained the etiology of his voiding and defecation disorders. It is important to examine the motivation for treatment. If the child is not motivated, the results of the treatment are much worse.

4.4.1.2 Behavioral modifications

The goal is to improve the control of urination and defecation through changes in the child's behavior. This includes regular voiding and hydration, taking the correct position during urination and defecation, as well as changing the diet. With the application of this initial treatment, a cure can be achieved in about 20% of children [21].

4.4.1.3 Regular hydration

Children with voiding disorders are often voluntarily dehydrated. Concentrated urine can cause irritation and burning when urinating. The introduction of adequate hydration (200 ml 5-6 times a day) increases urine production and bladder capacity, reduces urine concentration and improves bowel function. It is forbidden to drink beverages such as Coca-Cola, coffee, tea, cold juices, because they can induce detrusor overactivity [25].

4.4.1.4 Regular voiding

It is the basis of bladder rehabilitation and includes the adoption of a voiding schedule every 2-3 hours. The goal is to prevent bladder distension, to restore the feeling of fullness of the bladder, and to reduce the hyperactivity of the bladder. The child and parents should know that the ability to start micturition, even when the child does not want to urinate, is an important step in controlling the continence and emptying of the bladder. The child should try to urinate only at a predetermined time, and avoid delaying urination. It is necessary to analyze the child's daily activities and determine the time of micturition accordingly.

4.4.1.5 Optimal bladder emptying

The position when urinating has a significant effect on the ability to empty the bladder. Toilet bowls are intended for adults and as such are not suitable for children. Relaxation of the PFMs is difficult or impossible with various irregular positions that the child assumes during urination.

Proper voiding position means urinating in a sitting position on the toilet bowl for girls and boys. For smaller children, it is important to require the use of an adequate toilet seat insert and footrests to ensure trunk stability [26]. When urinating, the child should be slightly bent forward, with the spine in extension, the hips in abduction and the relaxed abdominal muscles (**Figure 3**). It is necessary for children to listen to the sound of the stream when urinating and the goal is for the stream to be strong, long and sonorous.

4.4.1.6 Treatment of chronic constipation/fecal incontinence

It is important to recognize the defecation disorder, because it has been proven that the treatment of constipation alone significantly reduces the symptoms of the lower urinary tract. In the group of children with increased post-void residual urine and constipation, 66% had an improvement in bladder emptying after constipation treatment. Urinary incontinence, nocturnal enuresis, and recurrent urinary tract infections were cured in most children treated only for constipation [20]. Therefore, treatment begins with chronic constipation management.

In the treatment of chronic constipation, four steps are applied: education, disimpaction of fecal mass, prevention of its re-accumulation and follow-up [27]. Treatment is usually applied for 3-6 months, but the relapses are frequent [28]. The cure rate of chronic functional constipation after application of standard treatment that includes laxatives and behavioral approaches is only 50-60% [29].

More than half of children with chronic constipation have an abnormal defecation pattern because they contract the external anal sphincter and the *M. puborectalis* during defecation [30]. This form of abnormal defecation is considered to be learned resulting from the habit of delaying defecations. Physiotherapy



Figure 3.
Correct position when urinating.

interventions such as diaphragmatic breathing exercises and pelvic floor exercises with or without biofeedback were introduced in order to educate a child to relax the external anal sphincter and the PFMs during defecation [31–35]. In refractory cases, even botulinum toxin injections are administered into the external anal sphincter [36].

Interferential current stimulation (IFS) has been used in the treatment of chronic constipation resistant to standard therapy in children. Significant improvements in clinical symptoms (increased frequency of defecation, reduction of fecal incontinence and abdominal pain) were seen in 67% of children and lasted for more than two years in one third of the treated patients [37]. In addition, the time of colonic transit on colonic scintigraphy was shorter after the application of IFS [38]. Although the mechanism of action of IFS is still insufficiently known, the proposed theories are the activation of local sensory nerves in the skin, spinal nerves (sensory and motor T9-L2), sympathetic and parasympathetic nerves in the intestine, enteric nerves, pacemaker cells (Cajal's interstitial cells) or smooth muscle cells in the intestinal wall [39].

5. Rehabilitation protocols for dysfunctional voiding

Although there is no general approach to treatment and treatment varies from one patient to another, there are several ways to treat children with DV, including urotherapy, pharmacotherapy, botulinum toxin application, and surgical treatment in children with VUR.

In addition to standard urotherapy, special measures are applied, such as exercises for relaxation of PFMs with or without biofeedback and exercises for relaxation of abdominal muscles.

5.1 Pelvic floor muscle relaxation exercises and biofeedback

Pelvic floor muscle exercises were first used in pediatric urology by Wennergren and Oberg, with the aim of developing the child's awareness of their function [40]. During the exercises, the child learns to contract and relax these muscles without activating auxiliary muscles (gluteal and hip adductors). In order to improve voluntary control, the exercises can be combined with different types of biofeedback, such as visual (observation of the abdomen in front of a mirror), tactile (palpation of the PFMs or *M. transversus abdominis*), uroflowmetry or electromyography.

Biofeedback was first used by Maizels et al. in 1979, who implemented the use of urodynamics devices in children with detrusor sphincter dyssynergia [41]. During urination, children observed EMG activity of the sphincter. Improvement was achieved in two of the three treated children.

Uroflowmetry biofeedback consists of the child observing the uroflowmetry curve while urinating. During voiding, the child is advised to make sure that the curve is bell-shaped. Kjolseth examined the efficacy of uroflowmetry biofeedback in 32 children with DV [42]. The number of applied sessions was 1-9, while 47% of children required 4-5 sessions. Cure was achieved in 50% of children, improvement in 8 children, and 7 children were unchanged. The uroflowmetry curve was completely normalized in 55% of children. It has been shown that this type of biofeedback requires a smaller number of sessions compared to EMG biofeedback and leads to faster normalization of the act of urination [43].

5.1.1 Animated pelvic floor EMG biofeedback

Mc Kenna et al. in 1999 applied biofeedback in the form of interactive computer games that enabled the active participation of patients [44]. Computer play maintained the child's interest and motivation for the exercise programme. The method consists of placing superficial EMG electrodes on the child's perineum, and then the child is taught to properly contract and relax the PFMs by watching a game on a computer monitor. In this way, children become aware of the activity of the PFMs and learn to control them by controlling the activities of their favorite heroes (dolphin, monkey, fish, bee).

In a study by Herndon et al. interactive computer games were used in 160 children with DV [45]. In 87% of patients, subjective improvement of symptoms was achieved. In a study by McKenna et al. improvement of nocturnal enuresis was achieved in 90%, daily wetting in 89%, constipation and fecal incontinence in 100% [44].

Kaye and Palmer did not find significant differences in efficacy after application of non-animated (biofeedback without animation using only EMG tracing) and animated biofeedback [46]. However, a group of children who had animated biofeedback required a smaller number of sessions to normalize the uroflowmetry curve and reduce residual urine. In a study by Desantis et al. there was an improvement in urinary tract infections in 83%, diurnal incontinence in 80%, constipation from 18-100%, urinary frequency from 67-100%, urgency from 71-88% and VUR from 21-100% of children [47].

In a study by Palmer et al. in children with DV and VUR, the use of biofeedback accelerated the resolution of VUR or reduced the degree of VUR in 71% of children [48]. Similar results were presented by Khen-Dunlop et al. and Kibar et al. [49, 50].

Adequate patient selection seems to be the most important for biofeedback success. Parents and children should be motivated and compliant to continue practicing exercises at home [51].

Although numerous studies highlight the positive effects of PFM relaxation exercises with or without biofeedback, there is no clear recommendation of an exercise protocol to use in the rehabilitation of children with DV. The number of sessions, the number of repetitions, the duration of the contraction and relaxation phase, as well as the period of performing the exercises differ significantly between the studies.

De Paepe et al. applied PFM relaxation exercises with EMG biofeedback [52]. The protocol consisted of 30 submaximal contractions lasting 3 seconds, followed by a relaxation phase of 30 seconds. One session per week was applied for 6 months (maximum 20-24 sessions). In a study by Vasconcelos et al., 24 home exercise sessions lasting 20 minutes were applied, three times a week during a three-month period [53]. The contractions lasted for 5 seconds, followed by a 10-second relaxation period. Shei Dei increased the duration of contractions to 10 seconds and extended the relaxation period to 30 seconds [54]. Yagci et al. applied submaximal contractions of 3 seconds, followed by a relaxation period of 30 seconds [55]. The children repeated the exercises at home three times a day for 6 months. In a retrospective study, Drzeviecki et al. analyzed a programme in which the contractions lasted 10 seconds, followed by a relaxation of 10 seconds [56]. After that, fast contractions lasting 5 seconds and 5 seconds of relaxation followed. On average, 3 sessions (1-8) were applied.

5.2 Abdominal capsule

Sapsford et al. showed that the PFMs are not an isolated unit, but a part of the abdominal capsule that surrounds the abdominal and pelvic organs [57]. The structures that make this capsule are the lumbar vertebrae, *M. multifidus*, diaphragm, *M. transversus abdominis* and PFMs. These muscles contribute to maintaining the posture of the body in an upright position and act synergistically.

Coactivation of the abdominal and PFMs is necessary for the development of intra-abdominal pressure and contributes to the stability of the spine. It is shown that *M. transversus abdominis* contributes the most to the development of intra-abdominal pressure in relation to other abdominal muscles [58]. This muscle is first activated during functions related to the increase in intra-abdominal pressure, such as spinal stabilization and expiratory tasks [58]. Coactivation of the abdominal capsule muscles has been demonstrated during weight lifting, coughing and forced expiratory tasks [58, 59].

Pelvic floor muscle dysfunction can present as hyperactivity, leading to the development of voiding and defecation disorders, such as DV, chronic constipation, perianal and perineal pain. Many of these children have hyperactivity of the lower abdominal muscles, which do not relax during urination and defecation and thus prevent the relaxation of the PFMs [57].

5.3 Diaphragmatic breathing exercises

As lower abdominal muscles (*M. transversus abdominis* and *M. obliquus internus abdominis*) and PFMs act synergistically, it is necessary for them to relax together during urination and defecation.

The simplest way for children to learn how to relax their abdominal muscles is through diaphragmatic breathing exercises. During diaphragmatic breathing, in inspiration, the diaphragm moves caudally and pushes the abdominal organs

forward. The anterior abdominal wall relaxes, as do the PFMs. This forward bulging of the anterior abdominal wall has been shown to reduce urethral pressure in healthy women and thus facilitate urination and defecation [58] .

Our institution was the first to incorporate this novel approach to treating DV. In a prospective clinical study of 43 children, in addition to standard urotherapy that included education on the importance of regular urination and hydration, proper voiding position and pattern, diaphragmatic breathing exercises and PFM relaxation exercises were performed in hospital settings for two weeks and then continued at home [60].

Diaphragmatic breathing exercises were performed in a supine position with the lower extremities supported by a pillow and hands placed on the abdominal muscles. The patient is required to inhale air through the nose, expel the anterior abdominal wall, hold the breath for a few seconds, and then exhale the air through the mouth (**Figures 4 and 5**). The exercises were repeated in both lateral positions, in the prone position, and then in the sitting position in front of the mirror (**Figure 6**). Children are required to observe the anterior abdominal wall during inspiration and then apply this exercise before urinating and defecating.

In addition, exercises for relaxation of the PFMs were performed. The child was placed in a lateral position with the upper leg flexed at the hip and knee and the lower leg extended. To enhance the proprioception of the PFMs, the examiner placed two fingers on the child's perineum and demanded that the child contract the PFMs without activating adjacent muscles such as the gluteus and hip adductor muscles. In this way, the child learned to localize and control the PFMs. The child was then required to perform submaximal contractions for 3 seconds followed by prolonged relaxation for about 30 seconds, for a total of 20 contractions. Children are required to perform these exercises daily at home for 6 months.

Control examinations were performed monthly for 12 months. Clinical manifestations (daytime urinary incontinence, nocturnal enuresis, urinary tract infections,



Figure 4.
Diaphragmatic breathing exercises in supine position (expiration).



Figure 5.
Diaphragmatic breathing exercises in supine position (inspiration).



Figure 6.
Diaphragmatic breathing exercises in front of the mirror.

constipation) were analyzed on a monthly basis and uroflowmetry was performed. The performance of diaphragmatic breathing exercises was controlled and the importance of daily exercise at home was emphasized. The children are encouraged to continue with the treatment.

After one year of monitoring and treatment, reevaluation of clinical manifestations and uroflowmetry parameters was performed. Urinary incontinence was cured in 83% of children, nocturnal enuresis in 63%, and urinary tract infections

in 68%. Chronic constipation was cured in all 15 patients. In addition, an objective improvement in uroflowmetry parameters was achieved. A normal uroflowmetry curve was registered in 90% of children.

The authors suggested that examination of lower abdominal muscles, recognition of their function during voiding and their relaxation should be incorporated in the treatment program of these children. Easy to learn diaphragmatic breathing exercises did not require any specific equipment and could be performed in children from five years of age. For centres that do not have access to pelvic floor EMG biofeedback, this programme could provide a treatment alternative as success rates are comparable to previous studies that used pelvic floor EMG biofeedback during urotherapy [54–56]. In order to achieve subjective and objective progress, children needed an average of 6.5 sessions, which is also equivalent to the average number of sessions in programmes that included non-animated biofeedback [46].

In the following study, the effects obtained in this group were compared with the effects in the group of children treated only with standard urotherapy (32 children) [31]. The children had 10 sessions of urotherapy in a hospital setting, and then were required to continue with it at home. After one year of follow-up, cure of urinary incontinence was achieved in only two children, nocturnal enuresis in 5, and urinary tract infections in 6 children. Constipation was cured in 6 out of 10 children. Uroflowmetry parameters did not show significant improvements. The authors concluded that diaphragmatic breathing exercises and PFM relaxation exercises, in combination with standard urotherapy, are important for the treatment of daily urinary incontinence, nocturnal enuresis and urinary tract infections, as well as for normalizing bladder function in children with DV.

5.4 Pharmacological therapy

Pharmacological therapy is considered an adjunct to improve bladder emptying in children with DV [43].

5.4.1 α -1 adrenergic receptor blockers

The role of α -1 adrenergic receptor blockers in the treatment of children with DV is controversial, as the mechanism of action at the level of the external urethral sphincter is still insufficiently known [61]. The possible mechanism of action is traditionally assumed to be relaxation of the periurethral, prostatic and bladder neck smooth muscles. In the study of Yucel et al., it has been shown that the effects of α -1 adrenergic blockers in reducing post-void residual urine can be compared with the effect of biofeedback [62].

5.4.2 Muscle relaxants

As DV is characterized by the inability of relaxation of the external urinary sphincter during urination, it was considered that muscle relaxant could be used in treatment.

Baclofen has been shown to be effective in reducing skeletal muscle spasticity, as well as in patients with striated sphincter dyssynergia [63]. However, the therapeutic effect is achieved only after the application of high doses. Serious adverse effects, especially after abrupt withdrawal, reduce its efficacy and safety in children with DV [64]. Therefore, tizanidine, a muscle relaxant used in many studies as a short-acting muscle relaxant due to spasmolytic action, was used. In a prospective, randomized study, 40 children with DV were divided into two groups [65]. The first group was treated with tizanidine (an imidazole derivative), while the

second group of children was treated with α -blocker (doxazosin). After 6 months of follow-up, both groups had similar improvement in symptoms and uroflowmetry parameters. In the doxazosin-treated group, urgency was the only symptom that showed a significant reduction after therapy. However, nocturnal enuresis, urgency, and daytime incontinence were significantly reduced in the tizanidine-treated group. Side effects were reported in 6 patients (15%). Epigastric pain was reported in two children (10%) receiving doxazosin. In the tizanidine group, loss of appetite was noted in two children (10%), epigastric pain in one (5%) and headache in one child (5%).

5.5 Botulinum toxin type A (BT-A)

BT-A is one of the strongest known toxins. When injected directly into a muscle, it causes flaccid paralysis by blocking the presynaptic release of acetylcholine [66].

The use of BT-A in patients with detrusor sphincter dyssynergia (DSD) was first described by Dykstra et al. [67]. In this study, BT-A was injected into the external urinary sphincter of adult patients with spinal cord injury and DSD. Positive results, reduced urethral pressure, and volume of residual urine remained for an average of 50 days.

Indications for the injection of BT-A in the external urinary sphincter have been extended over time to adult patients with DV and detrusor hypocontractility. In the study by Kuo et al. clinical and urodynamic improvement was registered in 83% of patients with urethral sphincter non-relaxation and detrusor hypocontractility [68]. Petit and co-workers reported a significant reduction in detrusor and urethral pressure, as well as the volume of post-void residual urine after a single injection of 150 units Dysport (BT-A) in patients with spinal cord injury and DSD [69]. The beneficial effects of the therapy lasted for about 2-3 months.

In children treated with BT-A (amp. Dysport) due to spasticity, the most commonly reported adverse effects were local muscle weakness, urinary incontinence, fatigue, somnolence, flu-like symptoms, fever, and rash [70].

BT-A is used in the treatment of DV in children who are resistant to standard urotherapy. In the study of Radojicic et al., BT-A was applied in to the external urinary sphincter in children with DV [71]. The residual urine decreased significantly in 17 of 20 patients after 6 months of follow-up. The authors emphasized that temporary inhibition of the external urinary sphincter and/or PFMs may interrupt the DV cycle. The use of urotherapy during this period could help the child to re-adopt a normal urination pattern and thus reduce the need for re-injections.

In our institution, a prospective clinical study included 9 neurologically healthy girls with DV, aged 3-11 years, who had previously been treated with standard urotherapy without improvement [72]. Application of BT-A (amp. Dysport) in a dose of 500 units was performed transperineally into the external urethral sphincter. After two weeks of application, rehabilitation treatment consisting of standard urotherapy and PFM relaxation exercises was included. Six months after Dysport administration, there was a statistically significant improvement in clinical manifestations (urinary incontinence, voiding difficulties, urinary tract infections), and a significant reduction in post-void residual urine. No significant improvement in uroflowmetry parameters was registered. No children had systemic side effects with Dysport. The authors concluded that the act of urination in children with DV resistant to standard therapy can be significantly improved and maintained for at least 6 months after the use of amp. Dysport and urotherapy.

5.6 Manual physical therapy

Manual physical therapy with an osteopathic approach (MPT-OA) entails palpation and receptive manipulation of body tissues to relieve restraints that limit mobility and health. Biomechanical, myofascial, and articular constraints can contribute to DV by altering alignment, distorting the pelvis, restricting mobility, and thus affecting pressures within the abdominal and pelvic cavities [73]. Altered pressure relationships can affect neurological, vascular, lymphatic, and hormonal functions. In the randomized controlled trial that involved 21 children with DV, it has been shown that children with additional 4 sessions of MPT-OA demonstrated better short-term results compared to children that had only standard treatment [73]. The authors speculated that MPT-OA treatment helped restoring more natural alignment and mobility which helped the abdominal and PFMs to function more efficiently. However this single-centre promising results should be confirmed by multi-centre randomized controlled trials in order to draw definitive conclusions of MPT-OA in children with DV.

6. Conclusion

Urotherapy is the cornerstone of DV care for children. The treatment begins with standard urotherapy, after which specific measures are added. Rehabilitation programmes with diaphragmatic breathing exercises and pelvic floor relaxation exercises are superior in curing lower urinary tract symptoms and normalizing urinary function than programmes that only include standard urotherapy. The success rates of programmes that included pelvic floor relaxation exercises and diaphragmatic breathing exercises without the use of pelvic floor EMG biofeedback were equivalent to those that included pelvic floor EMG biofeedback, proposing a treatment choice for centres that do not have access to EMG biofeedback. However, no standardized pelvic floor exercise protocol exists. Therefore, new prospective multicentric randomized trials with a larger number of children are needed to determine the most appropriate programme that will have the best therapeutic outcome.

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Conflict of interest

None declared.

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