Learned Voiding Dysfunction (Non-Neurogenic, Neurogenic Bladder) Among Adults

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Data concerning learned voiding dysfunction (Hinman syndrome; non-neurogenic, neurogenic bladder) in adults are scarce. The present study was conducted to assess the prevalence and clinical characteristics of this dysfunction among adults referred for evaluation of lower urinary tract symptoms. Learned voiding dysfunction was suggested by a characteristic clinical history and intermittent ''free'' uroflow pattern and by the absence of any detectable neurological abnormality or anatomic urethral obstruction. A definitive diagnosis was made by the demonstration of typical external urethral sphincter contractions during micturition by EMG or fluoroscopy. A urodynamic database of 1,015 consecutive adults was reviewed. Twenty-one (2%) patients (age, 24–76 years) met our strict criteria of learned voiding dysfunction. Obstructive symptoms were the most common presenting symptoms, followed by frequency, nocturia, and urgency. Eight (35%) patients had recurrent urinary tract infections, seven of these being women. None of the patients had any clinically significant upper urinary tract damage. First sensation volume was significantly lower in women than in men. Both detrusor pressure at maximum flow and maximum detrusor pressure during voiding were found to be significantly higher in men than in women. Further differentiation between adult women and men failed to reveal any other clinically significant differences. In conclusion, by strict video-urodynamic criteria, 2% of our patients had learned voiding dysfunction. Other patients, with presumed learned voiding dysfunction, who did not undergo video-urodynamics were not included in the present series. Thus, the prevalence of learned voiding dysfunction among adults referred for evaluation of lower urinary tract symptoms is likely to be even higher. *Neurourol. Urodynam. 20:259–268, 2001.*   © 2001 Wiley-Liss, Inc.

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INTRODUCTION

The non-neurogenic, neurogenic bladder, also known as the Hinman syndrome, is a functional bladder outlet obstruction caused by voluntary contractions of the external urethral sphincter during voiding. It is believed to be an abnormal, learned response to uncontrolled bladder contractions, which is usually developed early in life [McGuire and Savastano, 1984; Hinman, 1986]. It has been suggested that this incoordination of the micturition process is due to a learned behavioral disturbance and may be reversed

by re-educational therapy [Hinman, 1986]. We prefer the term *learned voiding dysfunction* to describe this condition.

Typically, children with a non-neurogenic, neurogenic bladder present with frequency, urgency, urinary incontinence, recurrent urinary tract infections, or occasionally, encopresis. Further evaluation may reveal signs of obstructive uropathy, such as trabeculated bladder, elevated post-void residual urine, hydronephrosis, and vesicoureteral reflux, in the absence of any identifiable neurological or obstructive abnormality. Urodynamically, these children have reflex detrusor contractions that they fail to inhibit, and they do not coordinate these contractions with concomitant sphincteric relaxation. In time, not only do they find it difficult to inhibit detrusor contractions, but they also find it difficult to keep the sphincter relaxed when voiding occurs.

In children, learned voiding dysfunction is usually acquired after toilet training, reaches its peak of destructiveness in late childhood, and tends to resolve after puberty [Allen, 1977]. Although it may persist, or even first manifest, later in life, data concerning learned voiding dysfunction in adults is scarce. The present study was conducted to assess the prevalence and clinical characteristics of this learned voiding dysfunction among adults referred for evaluation of lower urinary tract symptoms.

METHODS

A urodynamic database of 1,015 consecutive men and women, referred for evaluation of voiding symptoms over a period of 4 years, was reviewed. All patients underwent meticulous clinical evaluation, which included a complete history and physical examination, urinary questionnaire, voiding diary, pad test, urine culture, non-invasive uroflowmetry, post-void residual urine volume, and video urodynamics. All patients underwent renal ultrasound evaluation to exclude upper urinary tract abnormalities. Urethrocystoscopy was performed according to clinical indications.

Before examination, all patients voided in private using a standard toilet and the noninvasive (''free'') flow was recorded. Residual urine volume was measured by ultrasound examination immediately after bladder emptying. Multichannel video urodynamics were performed according to the recommendations of the International Continence Society [Abrams et al., 1988] except for cystometry. Contrary to the above recommendations, the patients were not instructed to try to inhibit micturition during the filling phase, but rather to report sensations to the examiner. The cystometrogram was performed using radiographic contrast and a 7-F double-lumen catheter via constant infusion at a medium filling rate, with rectal pressure monitoring. Perineal surface electrodes were used for electromyography (EMG). At capacity, patients were asked to void, and pressure flow studies with simultaneous video fluoroscopy of the bladder outlet and EMG activity were performed.

As no standard definitions for learned voiding dysfunction in neurologically normal adults have been established, we considered clinical evidence of this disorder as: 1) a suggestive clinical history, e.g., lower urinary tract symptoms and difficulty in voiding in public places, or during uroflowmetry/urodynamics, having to concentrate, relax, touch genitalia, listen to running water; 2) intermittent ''free'' uroflow pattern; and 3) neurological disorders, or anatomical causes of bladder outlet obstruction are excluded. This was determined by history, physical examination, urodynamics, and additional imaging studies, such as MRI, when results of the clinical evaluation raised

any suspicion of neurological or anatomical finding. A definitive diagnosis was made by the demonstration of typical external urethral sphincter contractions during micturition with needle EMG or fluoroscopic visualization of the urethra during voiding. Previous studies showed that contrary to detrusor-external sphincter dyssynergia (DESD), in which the onset of the detrusor contraction is preceded by an increase in sphincter EMG activity, in learned voiding dysfunction, the sphincter EMG activity diminishes just before the contraction. The activity then sporadically increases as the patient contracts and relaxes the sphincter [Blaivas et al., 1981a,b]. However, these observations were made with needle EMG and most probably cannot be made with surface EMG. If needle EMG is unavailable, the characteristics of the urethra seen at fluoroscopy often provide enough information for a definitive diagnosis. The pelvic floor−external sphincter complex can be observed to contract and relax during voiding. The urethra is usually dilated to the level of the external sphincter, while the bladder neck is wide open, distinguishing dysfunctional voiding from primary bladder neck obstruction. Typical video urodynamic findings are presented in Fig. 1.

Patient characteristics as well as urodynamic parameters were all analyzed and compared between women and men. Results were analyzed statistically by Student's t-test and χ^2 test. Values of $P < 0.05$ were considered significant. Data are presented as mean $\pm$ SD, or percentage according to the variables.

RESULTS

Incidence and Patient Characteristics

Of the 1,015 (620 women and 395 men) consecutive patients in the database, 21 (2%; 13 women and 8 men) met our criteria of learned voiding dysfunction. The mean age of the 13 women was 51.5 ± 9.2 years (range, 40–69). Five (38%) women, two of whom were on hormone replacement therapy, were postmenopausal. The mean age of the eight men was 44.9 ± 17.1 years (range, 24–76).

Symptoms and Physical Examination

Obstructive symptoms (e.g., hesitancy, weak or intermittent stream, or incomplete emptying) were the most common presenting symptoms (95% of the study population), followed by frequency (62%), nocturia (62%), and urgency (52%). Only four patients (19% of the study population) complained of urinary (urge and/or stress) incontinence. Eight (35%) patients had recurrent urinary tract infections, seven of these being women. Timing of onset of symptoms was often difficult to determine from history. However, 14 patients were available for up-to-date follow-up and a focused re-anamnesis. All denied any childhood problems related to the urinary tract.

Severity of the symptoms was assessed using the American Urological Association (AUA) symptom index score for benign prostatic hyperplasia. The total scores were classified as mild (0–7), moderate (8–19), and severe (20–35). Ten patients were found to have severe symptoms, while in all the others the symptoms were moderate (48 and 52% of the study population, respectively). None had mild symptoms. The mean AUA symptom index scores were similar among women and men (23.2 ± 7.9 and 26.4 ± 8.6, respectively).

All patients underwent focused pelvic examination. None of the women had any clinically significant urogenital prolapse and none had any palpable urethral diver-
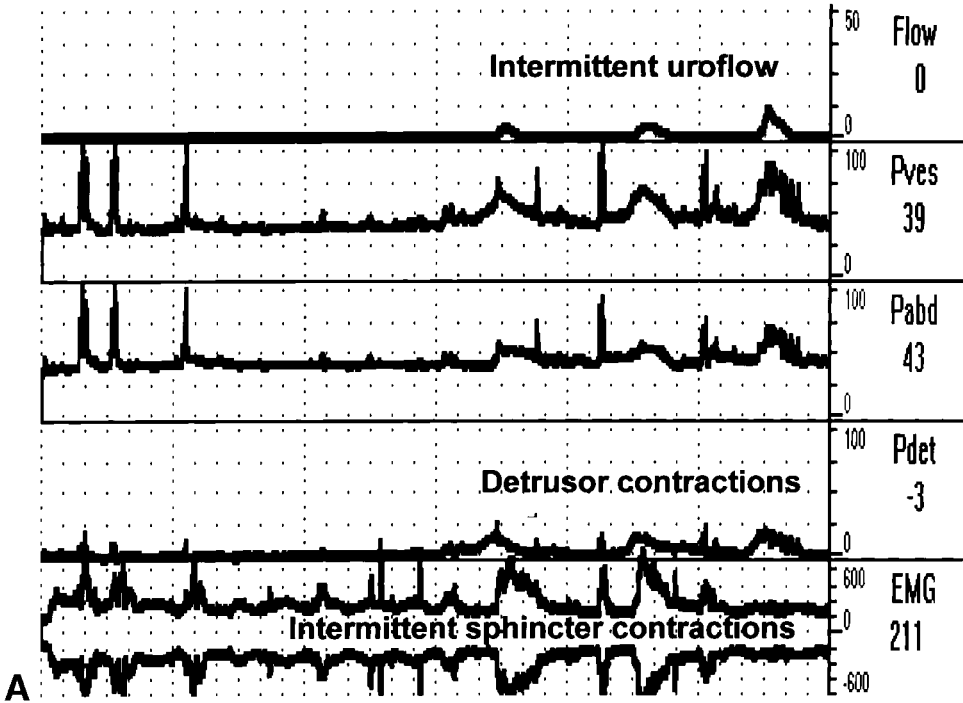
Fig. 1.   Learned voiding dysfunction. **A:** Typical urodynamic tracing. **B:** Fluoroscopic visualization of the urethra during voiding.

ticulum. Rectal examonation revealed only one slightly enlarged, tender, prostate in a 36-year-old man. All others had unremarkable findings.

Upper Urinary Tract Evaluation

None of the patients had any clinically significant upper urinary tract damage, e.g., hydronephrosis, stones, atrophy, or reflux. One patient had minimal (Grade 1) unilateral pyelocaliectasis.

24-Hour Voiding Diary

All patients completed a 24-hour voiding diary. The mean diurnal and mean nocturnal micturition episodes were almost identical in women and men (12.1 ± 6.8 vs. 12.0 ± 6.4; and 3.5 ± 3.6 vs. 3.0 ± 1.3, respectively). The mean largest voided volumes were similar as well (369 ± 134 vs. 310 ± 220 mL, respectively).

Uroflowmetry

The non-invasive (''free'') uroflow study was repeated at least twice to insure consistency. All patients were found to have a persistent intermittent free-flow pattern. The mean values of the peak flow rate, mean flow rate, flow time, voided volume, and post-void residual urine were similar among women and men (Table I).

Urodynamic Findings

First sensation volume was significantly lower in women than in men (123 ± 90 vs. 272 ± 128 mL, respectively. $P = 0.006$). A clear trend of lower values was also evident in all other cystometric parameters (first urge, strong urge, and bladder capacity) in women vs. men, although statistical significance was not reached (Table II). Three patients with learned voiding dysfunction were also found to have detrusor instability. Two other women had sphincteric incontinence.

Both detrusor pressure at maximum flow (pdet. Qmax) and maximum detrusor pressure during voiding (pdet.max) were found to be significantly higher in men than in women (Table II).

Further analysis was carried out to compare uroflow parameters in non-invasive (''free'' flow) vs. intubated (pressure-flow) uroflowmetry. A significant difference was found between maximum flow rate and residual urinary volume with regard to free uroflowmetry compared to intubated uroflowmetry (15.7 ± 7.9 vs. 6.4 ± 5.1 mL/s, $P = 0.0002$; and 58 ± 91 vs. 216 ± 238 mL, $P = 0.01$; respectively), although the mean voided volumes in both techniques were similar (250 ± 167 vs. 237 ± 167 mL, respectively).

TABLE I. Non-invasive Uroflow Parameters in Patients With Learned Voiding Dysfunction

Mean $\pm$ SD	Women (N $=$ 13)	Men (N $=$ 8)
Peak flow rate (mL/s)	15.6 ± 8.7	15.8 ± 6.6
Mean flow rate (mL/s)	8.8 ± 5.5	9.9 ± 4.6
Flow time (sec)	27.5 ± 20.1	39.6 ± 24
Voided volume (mL)	238 ± 187	269 ± 132
Post-voided residual urine (mL)	82 ± 107	28 ± 52

TABLE II. Urodynamic Parameters in Patients With Learned Voiding Dysfunction*

	Women (N = 13)	Men (N = 8)	*P* value
First sensation (mL)	123 ± 90	272 ± 128	0.006
First urge (mL)	224 ± 111	326 ± 160	0.08
Strong urge (mL)	337 ± 144	435 ± 177	0.12
Bladder capacity (mL)	429 ± 172	594 ± 296	0.08
Detrusor instability	2 (15%)	1 (13%)	
pdet. Qmax (cm H_2O)	19.8 ± 12.6	43.8 ± 11.5	0.002
pdet. max (cm H_2O)	31.5 ± 14.0	53.3 ± 14.8	0.005

*pdet. Qmax: detrusor pressure at maximum flow; pdet. max: maximum detrusor pressure during voiding.

Urethrocystoscopy

Fifteen patients (10 women and five men) underwent diagnostic urethrocysto-scopy. Cystoscopic findings in the 10 women were either unremarkable (50%) or benign (mild to moderate trabeculation, 40%; midurethral scarring, 10%). Cystoscopic evaluation of the five men revealed moderate to severe bladder trabeculation in all and bladder neck stricture or scarring in two subjects, both of whom previously had undergone TURP.

Management and Follow-Up

All our patients were advised to undergo behavioral modification; however, only five did follow through. Fourteen (61%) patients (eight women and six men) were available for up-to-date follow-up. Median duration of follow-up was 18 months (range, 6–38). Three women underwent behavioral modification, two of whom were significantly improved after therapy. Three women with mixed voiding dysfunction were managed, and partially improved, by different treatment modalities (antic-holinergics, collagen injections, or pentosan polysulfate sodium [Elmiron]). Two other women had no treatment, one reported spontaneous improvement, and the other was unchanged.

Two men underwent behavioral modification and both reported improvement. Of the four remaining men, two underwent bladder neck incision and reported improvement, one reported improvement with antibiotics and self-catheterization, and another one was unchanged and has sought no treatment.

DISCUSSION

The prevalence of learned voiding dysfunction among adults is unknown. Jorgensen et al. [1982] reported 0.5% prevalence rate among patients referred for urodynamic evaluation. Results of the present study suggest that this disorder is more common than previously recognized, occurring in 2% of our patients. Moreover, this database considered only patients who underwent video-urodynamic studies. Other patients, with presumed learned voiding dysfunction, who did not undergo urodynamics were excluded. Thus, the prevalence of learned voiding dysfunction among our patients is probably higher.

In 1971, Hinman presented 14 boys with typical characteristics of the non-neurogenic, neurogenic bladder syndrome, and introduced a new concept, i.e., that these changes were behavioral as demonstrated by their reversal by suggestion (including

hypnosis) and by the absence of any detectable neurological or obstructive abnormality [Hinman, 1971]. Since then, several studies have shown that these children have reflex bladder contractions that they fail to inhibit and concomitant urethral sphincter contractions that they fail to relax. With time, the child finds it difficult to keep the sphincter relaxed even during voluntary voiding [Allen, 1977; Koff et al., 1979; Jorgensen et al., 1982; McGuire and Savastano, 1984]. This incoordination of bladder-sphincter function eventually leads not only to lower urinary tract symptoms, but may cause structural and functional changes, including 1) obstruction and consequent bladder trabeculations, residual urine, and bacteriuria and 2) detrusor hypertrophy and consequent vesico-ureteral reflux or hydroureter/hydronephrosis [Hinman, 1986]. Hinman concluded, ''since these children usually are toilet trained initially, the incoordination appears to be a learned behavior or habit, perhaps as a response to under-appreciated detrusor contractions. Reversal of the syndrome is achieved by suitable medication and by some form of suggestion or retraining.'' Several synonyms of this syndrome have been suggested. Among these, the terms non-neurogenic, neurogenic bladder and Hinman syndrome gained wide popularity. We prefer to use the term learned voiding dysfunction because it reflects the underlying pathophysiology (i.e., learned behavior).

Clearly, a similar acquired malfunction may be responsible for some refractory lower urinary tract symptoms in adults. However, data concerning learned voiding dysfunction among adults are scarce. George and Slade [1979] reported a series of 16 men (mean age, 42 years; range, 29–55 years) referred for evaluation of refractory lower urinary tract symptoms. Their main symptoms were urinary frequency, hesitancy, intermittent stream, and inability to void in public places. These symptoms were found to be associated with a high incidence of dyspepsia and anxiety. The investigators suggested the existence of a chronic systemic state and proposed the term *anxious bladder*. Jorgensen et al. [1982] reported the symptomatology and clinical manifestations of ''idiopathic detrusor sphincter dyssynergia'' in neurologically normal patients referred for evaluation of voiding symptoms. Diagnosis was established by the following criteria: 1) two flow curves obtained in privacy showing a characteristic intermittent pattern and 2) simultaneous record of pressure-flow parameters and EMG demonstrating intermittent pelvic floor activity during micturition. Twenty-three patients (0.5% of the study population) fulfilled these criteria. The mean age of these patients was 27.4 years (range, 5–72). However, further differentiation between children and adults was not carried out. We used similar criteria to define learned voiding dysfunction, but also believe that if EMG is unavailable, the characteristics of urethral contractions seen at fluoroscopy often provides enough information for a definitive diagnosis. If EMG is available, concentric needle electrode provides a significantly superior signal source compared with the surface electrode.

Contrary to children, in whom the main subjective hallmarks of the syndrome are urinary incontinence and recurrent urinary tract infections, adult patients in our series had mainly obstructive and irritative symptoms, while urinary incontinence was less prominent. Recurrent urinary tract infections were more common among women than men (54 vs. 13%, respectively). Further differentiation between adult women and men failed to reveal any other clinically significant differences.

None of our patients had any evidence of upper urinary tract injury. This suggests that the learned behavior was too short in duration to cause significant damage. Moreover, all 14 patients whom we were able to contact for up-to-date follow-up denied

childhood problems related to the urinary tract. Had we been able to demonstrate that our adult patients were diagnosed with voiding dysfunction in childhood, the finding of an undamaged upper excretory system combined with low values for the maximum detrusor pressure and pressure at maximum flow might have suggested milder childhood disease in these subjects or decompensation of the detrusor muscle over time. Due to the incomplete information and the small number of cases, no speculations can be attempted at this time.

Optimal management of children with Hinman syndrome requires accurate, as well as timely, diagnosis. Hypnosis was the first modality successfully applied, in combination with anticholinergics and antibiotics [Hinman, 1986]. Over the years, suggestion, retraining, bladder drill, and biofeedback have been combined with pharmacologic therapy to treat detrusor instability, obtain striated muscle relaxation, or to inhibit contraction of the α-adrenergic innervated bladder neck. Although potentially successful in more than 80% of cases, the treatment can span 6 weeks to several years with occasional relapses, requiring cooperation and determination on the part of the child and her/his family [Hellstrom et al., 1987; Jerkins et al., 1987; Phillips and Uehling, 1993; Yang and Mayo, 1997]. Subjects with irreversible renal damage at the time of diagnosis and those who do not respond to, or drop out of behavioral therapy are candidates for reconstructive surgery, dialysis, or renal transplant. Reparative operations will fail if performed before diagnosis of dysfunctional voiding and optimization of the involved variables [Hinman, 1986; Yang and Mayo, 1997].

Data concerning the management of learned voiding dysfunction in adults are scarce. The correct diagnosis relies on detailed history corroborated by urodynamic studies with EMG or fluoroscopy, and may be missed due to inadequate awareness or instrumentation. It is, therefore, impossible to analyze series presented as "dysfunctional voiding" or "idiopathic." Recently, Deindl et al. [1998] suggested two different pathogenetic mechanisms of functional urethral obstruction in women with dysfunctional voiding and/or urinary retention. The activity patterns during micturition of both pubococcygeal muscles and the striated external urethral sphincter were assessed using two different EMG techniques. Four women were found to have inappropriate urethral sphincter activation during micturition. Eleven others had inappropriate contractions of the pubococcygeal muscles with no abnormality of the external sphincter. Biofeedback training led to improvement in women with pubococcygeal activation, but not for those with inappropriate urethral sphincter activation. Neuromodulation for patients who have failed numerous other therapies has been previously advocated but, so far, data are only anecdotal [Van Kerrebroek, 1998]. Most recently, Rosario et al. [2000] studied the potential contribution of ambulatory urodynamic monitoring in men with urinary symptoms unable to initiate a void on conventional video cystometrography ("the bashful bladder"). In 11 patients, a history of being unable to void at a public urinal was noted. Reassurance of normalcy, based on the ambulatory urodynamic results, was associated with spontaneous improvement in 42% of the patients. The authors concluded that the contribution of ambulatory urodynamic monitoring compared to more conventional investigations in men younger than 40 years is negligible, but the test may serve to reassure the anxious patient and help select appropriate nonsurgical therapy. In our series, five patients underwent behavioral modification, four of whom considered themselvs significantly improved or cured. Although the numbers are small and care should be taken in their interpretation, behavioral modification seems

to result in good short- and medium-term outcomes. Other patients in our series, with mixed voiding abnormalities, were improved following different treatments, or no treatment at all. This may impy that learned voiding dysfunction may be the end result of various pathophysiologic mechanisms. Further exposure of these mechanisms may enable the use of target, more effective, treatments. Larger series and long-term follow-up are required to establish the optimal treatment modality of this voiding dysfunction.

In conclusion, learned voiding dysfunction in adults appears to be more common than was previously recognized, occurring by strict video-urodynamic criteria in 2% of our patients. Other patients, with presumed learned voiding dysfunction, who did not undergo video urodynamics, were not included in the present series. Thus, the prevalence of learned voiding dysfunction among adults referred for evaluation of lower urinary tract symptoms is likely to be even higher.

Micturition symptoms relevant to learned voiding dysfunction are very suggestive but inconclusive, and a full urodynamic evaluation, including simultaneous EMG recording or fluoroscopy, is essential in making the definitive diagnosis and formulating a treatment plan. Timely diagnosis of this disorder may offer the best opportunity to reverse functional, as well as structural, abnormalities by non-invasive measures.

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