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Martin T. Stein, Madhu Alagiri and Daniel P. Kohen  
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# CHALLENGING CASE: CHRONIC DISEASE—DEVELOPMENTAL AND BEHAVIORAL IMPLICATIONS

## Diurnal and Nocturnal Enuresis in a 6 Year Old\*

### CASE

Justin is a 6-year-old boy diagnosed with attention-deficit/hyperactivity disorder who presented with daytime and night-time wetting. He had been toilet trained at the age of 3 years but was back in diapers as a result of the enuresis. His bowel movements were normal. The initial evaluation consisted of a urinalysis, urine culture, serum creatinine level, and renal/bladder ultrasound examination. The urine studies were normal. The serum creatinine level was elevated at 1.0 mg/dL. The ultrasound examination revealed bilateral mild hydronephrosis and a thickened bladder wall. A voiding cystourethrogram was ordered to evaluate anatomy, but Justin would not allow a Foley catheter to be inserted, so the procedure, along with cystoscopy, was performed under anesthesia. Cystoscopy revealed a highly trabeculated bladder, as is seen in either high-grade obstruction or a neurogenic bladder. The cystogram did not show any obstruction or vesicoureteral reflux. Meanwhile, Justin's symptoms continued to increase to the point at which he was constantly wet, and he no longer made any attempts to void on his own. A spinal magnetic resonance imaging study did not show any occult neurologic lesions. Urodynamic studies revealed a high-pressure bladder, poor emptying, and inappropriate voluntary contraction of the striated, urinary sphincter during micturition. Despite institution of anticholinergic medication, psychotherapy, and behavioral therapy, Justin continued to do poorly. He could not tolerate clean intermittent catheterization, and he eventually required a suprapubic tube for urinary drainage and preservation of his kidneys.

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Index terms: *diurnal enuresis, nocturnal enuresis, Hinman syndrome, non-neurogenic neurogenic bladder.*

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### Dr. Martin T. Stein

For a primary care or developmental-behavioral pediatrician on a general pediatric service, inpatient rounds can be challenging, rewarding, and enlightening. During my last assignment as an attending pediatrician, this 6-year-old child with Hinman syndrome was presented on rounds. The pediatric resident stated, in a manner that suggested a routine admission, that the child was "admitted by pediatric urology for a diverting cystostomy as a result of Hinman syndrome." I immediately pointed out my ig-

norance of this disorder. Subsequent discussions with the pediatric urology staff and a search of both the pediatric and urologic literature revealed a fascinating, complex, and potentially debilitating behavioral-urologic disorder of childhood.

Hinman syndrome is also known by the seemingly contradictory term, *non-neurogenic neurogenic bladder*. As a result of a chronic process of voluntary contraction of the urinary sphincter, these children, who have structurally normal urinary tracts, develop a bladder that functions like an upper motor neuron-type of neurogenic bladder. Although diurnal and nocturnal enuresis might be the only symptoms in a mild case, an obstructive uropathy and renal failure might ensue in the more severe cases. That the original description of the syndrome was reported by a urologist (F. Hinman) and a pediatrician (F.W. Baumann) is significant, in that the leading hypothesis for the pathogenesis of the disorder is a dysfunction of micturition initiated by psychological stress during early childhood. These issues will be explored by a pediatric urologist and a developmental behavioral pediatrician.

**Dr. Madhu Alagiri** is an assistant clinical professor of urology at the School of Medicine, University of California, San Diego, at which he is currently the director of pediatric urology services. He was the attending urological surgeon for the child described in this Challenging Case. I am indebted to his assistance in guiding my understanding of the condition. **Dr. Daniel P. Kohen** is an associate professor in the Department of Pediatrics and Family Practice and the Department of Community Health, School of Medicine, University of Minnesota, at which he is the director of the Behavioral Pediatrics Program. Dr. Kohen has contributed extensively to the literature in the field of hypnotherapy for several common pediatric behavioral disorders. He is the coauthor of *Hypnosis and Hypnotherapy with Children*.

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Childhood voiding problems encompass a variety of disorders, from simple night-time enuresis to more complex daytime incontinence and inappropriate urine holding. Hinman syndrome, also known as *non-neurogenic neurogenic bladder*, represents an uncommon and extreme form of dysfunctional voiding.

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First described in 1973, the disorder, found mostly in boys, is characterized by the inappropriate voluntary contraction of the striated urinary sphincter during the process of micturition.<sup>1</sup> This results in a functional urinary obstruction that over time causes urinary tract infections, detrusor failure, hydronephrosis, and, eventually, renal insufficiency. In addition, inappropriate anal sphincter activity might also be present, resulting in chronic constipation or encopresis.

The exact etiology of the disorder is uncertain, but it is considered a behavior that is acquired during potty training, when children learn to control their urinary tract by actively contracting their urinary sphincter.<sup>2</sup> Bauer et al<sup>3</sup> postulated that this behavior becomes habitual when the child fails to differentiate between voluntary and involuntary voiding. As a consequence, urinary sphincter activity is maintained at all times. Family dynamics also play a role in the etiology. Allen and Bright<sup>4</sup> demonstrated that parents of children with Hinman syndrome, especially the father, tend to be domineering and intolerant of weakness or failure. Furthermore, wetting is seen as defiant behavior that should be punished.

Evaluation of the child suspected of having Hinman syndrome requires a thorough history and physical examination, as well as radiologic and urodynamic assessment. Simply watching the child void might provide clues to the problem. If the child strains or grunts and an intermittent stream is evident, then it is likely either an anatomic or functional obstruction is present. Radiologic imaging with an ultrasound examination and a voiding cystourethrogram will rule out obstructive lesions and typically reveal a large capacity, highly trabeculated bladder with a large urine residual. One-half of these patients will also have severe vesicoureteral reflux.<sup>3</sup> Spinal imaging with a magnetic resonance image study will rule out neurologic lesions. Urodynamic studies will objectively document a poorly compliant bladder and failure of the sphincter to relax once micturition begins.<sup>5</sup> When voiding does occur, it is often intermittent and accomplished only with the aid of the Valsalva maneuver.

Management of Hinman syndrome is predicated on the need to maintain good bladder and fecal drainage, prevent infections, and preserve the kidneys. The initial course of action is bladder and bowel retraining. This includes a high-fiber diet and strong encouragement to void frequently. Punishment for wetting is stopped, and good behavior is rewarded.<sup>6</sup> Prophylactic antibiotics and anticholinergics are instituted to prevent recurrent infections, decrease bladder instability, and reduce intravesical pressures.

Unfortunately, many of these conservative measures fail, so more aggressive therapy, such as clean intermittent catheterization and enemas, is warranted to ensure adequate urinary and fecal drainage. Meanwhile, bladder retraining is continued, and psychotherapy and biofeedback should be considered if they have not already been instituted. Occasionally, even these therapeutic interventions fail to prevent infections and renal deterioration. The next

step is to bypass the sphincter by surgically diverting the urinary stream. This ranges from a simple suprapubic tube to a more permanent device, such as a vesicocutaneous conduit using the appendix or a complete bladder diversion using an ileal conduit.

Effectively managing a child with Hinman syndrome can be challenging, and it often requires a multidisciplinary approach and involved parents. Once encountered, it leaves a lasting impression on both pediatrician and pediatric urologist alike.

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#### Dr. Daniel P. Kohen

Hinman syndrome, also known as *non-neurogenic neurogenic bladder* or *Hinman-Allen syndrome*, was originally described by Hinman and Baumann in 1973.<sup>1</sup> It has been reported infrequently during the past 25 years. The confusing nomenclature derives from the identification of a bladder that behaves like a neurogenic bladder in the absence of an identifiable causative neurologic lesion: "Unstable bladder may activate the sphincteric guarding reflex, leading to dyscoordination between the sphincter and the detrusor, and eventually overdistension of the bladder, with a reduced contraction power of the detrusor, and ultimately development of the equivalent of a neurogenic bladder."<sup>2</sup> This phenomenon has also been referred to as *external sphincter dyssynergia*,<sup>3</sup> and it results in dysfunctional voiding.

Appropriate and timely assessment and management of dysfunctional voiding is essential for early diagnosis and for avoidance of the significant morbidity associated with Hinman syndrome. Dysfunctional voiding resulted in severe morbidity in 11 (40%) of 27 patients able to be studied. Ten of the 11 patients underwent 17 surgical procedures;<sup>3</sup> they experienced outcomes similar to those experienced by 6-year-old Justin.

Justin's initial presentation of daytime and nighttime wetting is not particularly unusual or uncommon for a 6 year old, especially for a child with a concurrent diagnosis of attention-deficit/hyperactivity disorder. The initial evaluation included a review of the toilet training history, assurance of the absence of constipation as a common contributing cause of enuresis, and preliminary urine studies. When the renal-bladder ultrasound examination revealed ab-

normalities, additional studies were indicated. The functional neurogenic bladder was revealed through the presence of a highly trabeculated bladder in the absence of obstruction.

In the recent article by Yang and Mayo,<sup>3</sup> eight patients with Hinman syndrome were cured using nonoperative conservative treatments. As long ago as 1977, bladder retraining methods resulted in better outcomes than did surgical methods in modulating this functional disorder caused by a dyscoordination between detrusor contraction and sphincter relaxation.<sup>4</sup> In 1987, Hellstrom et al<sup>5</sup> reported on a 3-year follow-up of seven patients with functional non-neurogenic bladder dysfunction. They found that after treatment with bladder training and biofeedback, 51% had completely normalized voiding patterns by 1 year, and 76% had normalized patterns by 3 years.

A variety of behavioral methodologies have been effective in the management of voiding disorders. Although self-regulation strategies have been widely and effectively applied to the management of nocturnal enuresis,<sup>6,7</sup> less has been written concerning their application in the management of diurnal enuresis and severe forms as experienced by Justin.

Children with enuresis (nocturnal or diurnal) benefit substantially from first receiving a developmentally specific education concerning their genitourinary anatomy and physiology. We teach them about the heart as a pump that sends blood around the body, the kidneys that act as the "washing machine for the blood" and as the urine or "pee" factory, and the bladder that stores up the "pee" until it's time to "go." The brain is described as a part of the body that sends and receives the messages and signals that know when it's time to hold the urine in (by keeping the urine "gate" closed) and when it's time to urinate in the toilet by first signaling the body to walk to the bathroom and then to open the bladder gate to let the urine out in the toilet where it belongs. In creating a new, user-friendly drawing of the body for each patient and pacing the teaching of that drawing to the individual and developmentally specific needs and abilities of the child, we not only seek to develop the personal rapport essential to a positive therapeutic outcome but also to begin to offer the message to each child that we think that they *can* develop the control necessary to achieve the desired outcome of becoming dry.

In addition, this form of brain-bladder communication might be reinforced through variably structured training in self-hypnosis, in which the child learns to "give instructions to your brain and bladder to talk with each other."<sup>7</sup> Various forms of biofeedback might also have been useful for Justin and children with similar problems. With electromyographic feedback, specifically the type with computer game graphics, children are able to see and enjoy on a computer screen the immediate effect of even subtle changes in muscle contraction and relaxation. This format provides an easily understood and accepted metaphor for control that might then be appropriately reinforced through absorption, repetition, education, and concurrent behavior modifica-

tion techniques, such as calendars recording dry pants, days, or nights.<sup>8,9</sup>

In addition to the use of relaxation/mental imagery (self-hypnosis) and computerized biofeedback, some investigators (J.B. Reaney, personal communication) had positive outcomes when adapting various versions of the alarm systems designed for nighttime, to use during the day by children with daytime wetting. The alarm device functions as a biofeedback device, signaling the child at the first drop of "accidental" urination. The anticipation of such future signals presumably functions in a manner equivalent to conditioning, with a goal of preventing more wetting. As with most therapeutic approaches, effectiveness is enhanced when these signals are used concurrently with positive suggestion, education concerning the body, and supportive counseling/psychotherapy, as needed.

On the basis of clinical history and examination, urodynamic findings, and the patient's social situation, there is, at present, no effective way to definitively identify which children with Hinman syndrome will progress to having a severe complication.<sup>3</sup> Children being evaluated for diurnal and/or nocturnal enuresis should have dysfunctional voiding/Hinman syndrome on the list of differential considerations. At present, there is no consistent recommendation that all children with diurnal enuresis should have urodynamic studies. In view, however, of the morbidity experienced by children such as Justin, it seems appropriate to consider urodynamic studies if initial behavioral/self-regulation training interventions as described and/or time do not result in reasonably prompt improvement in symptoms.

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**Dr. Martin T. Stein**

There are other examples of acquired disorders associated with chronic and progressive contraction of striated muscle leading to physiologic dysfunction in children and adolescents. Tension headaches, muscular torticollis, and quadriceps contractures in some athletes are a few examples found in pediatric practice. The most common visceral form of these disorders is seen in children who voluntarily withhold stool, leading to chronic constipation and a tight, constricted anal sphincter. Dr. Alagiri noted that, unlike Justin, many children with Hinman syndrome also have constipation and encopresis. In addition, it is intriguing to speculate that pyloric stenosis, an acquired pediatric disorder of gastric outlet obstruction secondary to muscle spasm, might be in the same category.

By 6 years of age, most children achieve night-time bladder control. Maturation of the urinary sphincter function and dry nights occur in approximately 75% of children by age 4 years and in approximately 90% by age 8 years.<sup>1,2</sup> Because of this developmental pattern, a diagnosis of nocturnal enuresis is usually not considered until after 6 years of age. By comparison, daytime dryness is achieved by 50% of children at 2.5 years and by 90% of children at 4 years. At 6 years of age, Justin's chief complaint was diurnal and nocturnal enuresis. Appropriately, a primary care evaluation was initiated when the clinician recognized that the diurnal presentation was a red flag.

Daytime wetting might reflect increased bladder irritability (e.g. infection, a mass impinging on the bladder as in severe constipation, or, rarely, a bladder tumor), a structural abnormality, or vaginal reflux seen in overweight girls who experience daytime wetting secondary to urinating with their legs together, leading to reflux into the vagina.<sup>3</sup> "Giggle incontinence" is associated with daytime wetting after laughter in susceptible girls. A child's individual temperament might be an important factor. Some children with particularly short attention spans and who are easily distractible might continue an activity and not respond to the sensation of a full bladder. Although there is no evidence to support a predictable association between nocturnal enuresis and psychopathology, persistent diurnal wetting warrants a review of environmental stress in the home, school, and community.

Significantly, in Justin, the urinalysis and urine culture were normal. An abnormal urine/bladder ultrasound examination and a mildly elevated serum creatinine level triggered an urologic evaluation. This experience points out the importance of including an ultrasound examination study of the genitourinary tract and renal function assessment in a school-age child with both diurnal and nocturnal enuresis not explained by history, physical examination, urinalysis, or urine culture.

In the hospital, Justin was a quiet, reserved child who was difficult to engage in play or conversation. Additional exploration of his developmental and family history uncovered important information.

Justin was sent to live with an aunt at the time of his second birthday. This change of caretaker was a result of his mother's inability to care for him, in part related to maternal drug abuse. Toilet training was unsuccessful between 2 and 3 years of age. His aunt's frustration resulted in repetitive periods of verbal abuse when he was resistant to training. On several occasions, he was locked in a closet when he refused to use the toilet. Although bowel control was achieved by age 3 years, an extended period of bladder control did not develop. After his hospitalization at 6 years old and after several months of urinary drainage by a suprapubic tube, the tube was removed because of recurrent urinary tract infections. Justin gradually showed a response to behavioral therapy. Cognitive and behavioral counseling for Justin and his family, with an emphasis on teaching strategies for self-regulation of voiding, were continued.

Many children, unfortunately, experience delays and regressions in bowel or bladder control associated with verbal or physical abuse by a parent or other caretaker. Most of these children exhibit a form of resiliency, in that permanent sequelae of elimination functions do not occur. Specific behavioral or neuromuscular physiologic factors, which predispose some children to what Dr. Alagiri described as an inability to differentiate between voluntary and involuntary voiding and result in chronic contraction of the urinary sphincter, elude current knowledge. It seems reasonable to suggest the hypothesis that Hinman syndrome is the result of a combination of a susceptible child temperament (inattention to bladder sensory signals), a hyper-responsive urinary sphincter, and a negative reinforcement pattern of the toilet training.

Justin exhibited the most severe form of Hinman syndrome, which progressed to a functionally obstructive uropathy. Pediatric urologists evaluate many other children with an early, mild form of the same disorder (M. Packer, personal communication). These children present with diurnal enuresis, and at this point, the news is good. As carefully outlined by Dr. Kohen, a combination of developmentally specific education concerning genitourinary tract anatomy and function, coupled with instruction in self-regulation strategies directed at normal micturition, can result in decreased sphincter tone and resolution of the daytime wetting pattern. I suspect that most young children who wet during the day learn to control urination on their own! Clinicians who care for children can identify those with persistent symptoms and offer counseling and behavioral therapy.

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