High-intensity, short-term biofeedback in children with Hinman’s syndrome (non-neuropathic voiding dyssynergia)

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Abstract
Objective: To evaluate the long-term response to high-intensity, short-term biofeedback in children with severe voiding dysfunction.

Patients and methods: We retrospectively reviewed patients who underwent short-term, high-intensity biofeedback therapy from 1996 to 2004. Improvement was classified based on clinical and radiographic findings. Patients were categorized as having Hinman’s syndrome when, in addition to urinary incontinence, at least four of the following categories were present: sphincter dyssynergia, bladder trabeculation, large post-void residual (PVR), hydronephrosis, vesicoureteral reflux (VUR) and urinary tract infections. There were 14 patients (eight males and six females), 13 of whom had Hinman’s syndrome. Age when biofeedback was initiated varied from 5.6 to 12.9 years (μ = 8.9 ± 2.2). Before biofeedback, all had large PVRs, bladder trabeculation and sphincter dyssynergia. Nine had hydronephrosis and five had VUR. One patient had renal failure.

Results: Before biofeedback, the mean PVR was 109 ml (25–270 ml); after biofeedback, this decreased to 21 ml (0–150 ml), including two patients who eventually failed treatment. All 14 patients were able to relax their external sphincter and reduce the PVR during biofeedback and on short-term follow up. Long-term follow up (μ = 59.4 months) in 12 patients established that seven had a durable response with remission of symptoms, reduced PVR and radiographic improvement. In three, symptoms partially recurred over time and two failed treatment completely.
Conclusion: Short-term, high-intensity biofeedback achieves a durable response in the majority of children with Hinman’s syndrome. Long-term follow up is needed to assure compliance.

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Introduction

Voiding patterns and acquisition of bladder control in healthy children are age and maturation dependent. Although the neurophysiological process by which children acquire urinary control is not completely understood, various developmental stages are recognized. Conscious sensation of bladder fullness is expected between the first and second year, and the ability to control voiding commonly develops in the second to third year of life [1,2]. A normal voiding cycle requires a bladder that expands easily during the filling phase and then contracts to empty completely during the voiding phase. Premature detrusor contractions or significant increases in bladder pressure should not occur during filling. Bladder fullness triggers a detrusor contraction, followed by sphincter relaxation, which can be inhibited at will for a certain interval. During the normal voiding phase there should be complete relaxation of the external urethral sphincter muscle so the urine can be completely and smoothly released from the bladder, without interruption.

Voiding dysfunction is caused by an abnormality in one or both phases of the micturition cycle. Consequences may be urinary incontinence, incomplete emptying, and interrupted or intermittent flow of urine. Dysfunctional voiding disorders are divided into neuropathic disorders, which are clearly related to identifiable neurological conditions, and non-neuropathic or functional disorders. Voiding disorders in neurologically intact children are still not completely understood, but usually represent a medically benign although socially detrimental condition. The magnitude of these conditions may vary, and they can be classified according to their impact on the upper urinary tract into three main categories: mild, moderate and major dysfunctional disorders [3]. Included among the major dysfunctional disorders is Hinman’s syndrome.

Described by Hinman and Baumann [4] in 1972, Hinman’s syndrome is a rare severe dysfunctional disorder associated with voiding disturbances and upper urinary tract damage in the absence of anatomical obstruction or neurological problem. Other terms that have been used to describe this voiding disorder include non-neurogenic neurogenic bladder and non-neuropathic voiding dyssynergia [5–7]. It is characterized by clinical and urodynamic evidence of involuntary bladder outlet obstruction at the level of the external sphincter. Although the etiology of this severe condition remains unclear, it has been related to emotional, behavioral and psychiatric disturbances, and has been predominantly described in boys [5]. Children with Hinman’s syndrome manifest overactivity of the external sphincter/pelvic floor while being unable to inhibit detrusor contractions. Eventually these children develop the consequences of bladder outlet obstruction with elevated intravesical filling and voiding pressures with increased post-void residual urine (PVR). Ultimately, these children may develop a decompensated or non-compliant bladder with secondary hydroreteronephrosis in approximately two-thirds of patients and significant VUR in about half.

Patients with Hinman’s syndrome present with urinary incontinence associated with chronic urinary retention. Related symptoms are an intermittent or staccato urinary stream, elevated PVR, recurrent UTIs, fecal retention and encopresis. Uroflowmetry will confirm an intermittent voiding pattern with reduced flow rates and prolonged voiding times. Further urodynamic evaluation typically reveals detrusor instability and pelvic floor overactivity during filling and voiding, often associated with decreased bladder compliance and high voiding pressure. Radiological abnormalities vary from bladder trabeculation to uretero-vesical junction obstruction or reflux associated with dilatation of the upper urinary tract and renal damage. Treatment is aimed at improving the child’s ability to empty the bladder and bowel, and at preventing upper tract deterioration. The aim of this study is to report our short-term and long-term experience with high-intensity, short-term biofeedback in children with severe voiding dysfunction (Hinman’s syndrome).

Patients and methods

We retrospectively reviewed 14 patients, eight males and six females, diagnosed with severe voiding dysfunction who underwent high-intensity,
short-term biofeedback therapy from 1996 to 2004. Age when biofeedback started varied from 5.6 to 12.9 years ($\mu = 8.9 \pm 2.2$ years). Patient records, imaging studies and urodynamic evaluations were reviewed. Imaging studies included pre- and post-void renal sonograms, contrast voiding cystograms and renal scans, when appropriate. All patients were evaluated with an MRI of the spine to exclude spinal cord pathology. High-intensity, short-term biofeedback therapy was recommended in a highly select group of patients with severe voiding dysfunction, including 13 (93%) with Hinman’s syndrome. Patients were categorized as having Hinman’s syndrome when, apart from urinary incontinence, at least four of the following six categories were presented: UTIs, large PVR, sphincter dyssynergia, bladder trabeculation, hydronephrosis and VUR.

Before biofeedback, all patients were noted to have sphincter dyssynergia and bladder trabeculation, and 13 (93%) had diurnal urinary incontinence. The average PVR was 109 ml (25–270 ml). Ten patients (71%) presented with repeated episodes of UTI. Nine patients (64%) had hydronephrosis and five (36%) VUR. One patient presented with renal insufficiency. Constipation was an associated symptom in 10 patients (71%).

After the technique had been discussed and consent given by patient and family, cystourethroscopy was performed under general anesthesia to allow the percutaneous suprapubic placement of a Dewan dual-lumen catheter and to confirm the absence of anatomical urethral obstruction. After a resting period of 24–48 h, a cystometrogram/electromyogram (CMG/EMG) urodynamic evaluation was performed via the Dewan catheter, using perianal patch electrodes. The EMG bladder-sphincter dyssynergia pattern was demonstrated to the patient and family. All of the patients were found to have some degree of detrusor overactivity. Multiple sequential bladder fillings were performed in a single day, stressing the need to relax the sphincter during voiding while monitoring the patient’s EMG response and measuring PVRs. The main goal of each session was to re-educate the patient to relax the sphincter while voiding. Bladder cycling was repeated as many times as needed, often up to 10–15 fillings, until the patient was able to completely relax the sphincter during the voiding and empty the bladder to completion. Home monitoring of PVRs by the parents was continued for the following week, via suprapubic tube. The patient then returned for follow-up analysis of the PVR diary. Urodynamic evaluation was repeated to re-assess the voiding pattern. Based on the response, the catheter was removed or the session repeated. Complementing the treatment, a bowel program and anticholinergic therapy were recommended, when appropriate.

Short-term (up to 1 year) and long-term (up to 8 years) responses were analyzed, based on the clinical and radiographic outcomes. Assessment include urinary continence, urinary infections, constipation, hydronephrosis, VUR, bladder trabeculation and PVR, monitored both during the first week via suprapubic tube and subsequently with pre-/post-void sonograms at 6–12-month intervals. Improvement was classified based on clinical and radiographic findings.

**Results**

All 14 patients were able to relax their external sphincters and reduce their PVR during and shortly after the biofeedback sessions. All patients responded to one or two sessions separated by a week of home monitoring of PVR by the parents via the suprapubic catheter. Improvements in clinical and radiographic outcomes after biofeedback are illustrated in *Figs. 1 and 2*. Urinary incontinence, number of episodes of UTI and constipation reduced significantly after treatment. After biofeedback, the average PVR decreased from 120 ml to 21 ml (*Fig. 3*). If the two patients who failed treatment are excluded, average PVR among good and partial responders was 12 ml. The number of patients with bladder trabeculation, hydronephrosis and VUR also diminished considerably (*Fig. 4*). Long-term follow up (from 22 to 104 months) was based on the response in the PVR diary. Urodynamic evaluation was repeated to re-assess the voiding pattern. Improvement was classified based on clinical and radiographic findings.

**Figure 1** Clinical symptoms before and after treatment with high-intensity, short-term biofeedback therapy (BIOF) in children with severe voiding dysfunction. Fourteen patients were analyzed, 13 with Hinman’s syndrome. Of 13 patients who presented severe urinary incontinence before biofeedback, 11 (85%) improved after treatment. Ten patients initially had UTI episodes and six (60%) were cured. Constipation improved in 80%.
90 months, \( m = 59 \) months) was achieved in 12 (86\%) patients, six males and six females. Two female patients, aged 6.6 and 11.9 years, failed treatment. The elder one has significant developmental delay. She improved during the short-term evaluation, but symptoms returned after 2 years. Both patients had persistent urinary incontinence, UTI, constipation and an average PVR of 150 ml on long-term follow-up. Ten patients (83\%) sustained a long-term positive response to biofeedback, with clinical and radiographic improvement. During a mean period of 59 months, all 10 patients maintained a low PVR (\( \mu = 12 \) ml, compared to \( \mu = 120 \) ml before treatment). Among them was the 6-year-old patient who had renal insufficiency when he started biofeedback. At that time, he had bilateral hydronephrosis and a very trabeculated bladder, with a PVR of 240 ml. After 67 months of follow up he sustained the clinical and radiological improvements, with almost no PVR.

In three of the 10 patients who responded long term, some symptoms gradually recurred. The first, a 12-year-old female with hydronephrosis and grade IV VUR before biofeedback, developed recurrent episodes of UTI. She was treated with a ureteral reimplant and is doing well, with a 64-month follow up. Symptoms of daytime and nighttime incontinence reappeared in the second patient, who was 9 years old when treatment started. Although he was able to improve again after biofeedback retraining, he was immature and unable to comply with therapy for a longer period. Therefore, after 30 months of follow up, he was treated with Botox sphincter injections with only moderate results. The third was an 8-year-old female, who presented with bacteriuria and urinary incontinence after 3 years of follow up. The voiding and bowel programs were reinforced during consultations and her continence improved. She had no bladder trabeculation and had a small PVR after 80 months follow up. The other seven responders (58\%) maintained a full long-term response.

**Discussion**

The management of children with Hinman’s syndrome is difficult [6]. Hinman divided the management of this condition into four categories: (1) suggestion therapy, including hypnosis; (2) retraining and bladder drill (timed voiding); (3) drug administration; and (4) biofeedback [7]. Most patients require a combination of these measures. Currently, initial treatment includes antibiotic prophylaxis for infection, intensive bowel management with enemas and/or laxatives followed by long-term use of stool softeners and fiber supplementation, a strict timed and multiple voiding schedule, and anticholinergics to reduce detrusor instability. Pharmacological efforts to relax the striated muscle sphincter (e.g., diazepam) and smooth muscle sphincter (alpha-adrenergic blockade) have met with anecdotal success, as have psychotherapy and hypnotism [8–12]. Biofeedback therapy may be particularly effective in older
individuals when they can be made to appreciate the necessity and technique for relaxation of the voluntary pelvic floor musculature during the bladder contraction [13–15].

Although our approach of short-term, high-intensity biofeedback to treat this select group of difficult patients involves a more invasive technique requiring general anesthesia for initial placement of the Dewan percutaneous suprapubic dual-lumen urodynamic catheter, it offers several advantages. These include the ability to perform urodynamic evaluation in sensate children via the suprapubic catheter without the need for urethral catheterization, which in some children will produce artifactual contraction of the external urethral sphincter during the voiding phase of the
study. This approach also affords the opportunity for multiple sequential treatment cycles during 1 day, allowing for a more rapid, condensed period of treatment and a shorter learning curve for the patient and family. Rather than taking several weeks and multiple follow-up biofeedback sessions, this approach usually only required one full day of biofeedback therapy to achieve successful results. Furthermore, suprapubic measurements of PVR during the follow-up week are more reliable and provide for ongoing feedback at home, making the patient and family more confident of the results. It is important to stress that these patients still need long-term follow up to reinforce the therapy, to insure compliance with ancillary measures including timed voiding and a bowel regimen, and to monitor for the early recurrence of symptoms, infections, and/or elevated PVRs.

Conclusion

Short term, high intensity biofeedback achieves a rapid and durable response in the majority of children with Hinman’s syndrome. Long-term follow up is needed to assure compliance and long-term success.

References


