

Pediatrics

Bladder acontractility after hypospadias repair

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Introduction

Hinman and Bauman brought attention to an entity that causes unilateral or bilateral hydronephrosis, vesicoureteral reflux, and trabeculated or irregular bladder walls.¹ First referred to as non-neurogenic neurogenic bladder, the disease is a detrusor hyperactivity that patients try to suppress by increasing the activity of the external sphincter. Later the entity became known as Hinman Syndrome. The name was first improperly used, not discriminated, for different voiding dysfunction conditions which led the International Children's Continence Society to reserve the name "Hinman Syndrome" only for more severe cases, of relevant consequences for the patient's renal function.²

Voiding dysfunctions, particularly in the presence of a vesicosphincterian dyssynergia, may develop into hydronephrosis and renal insufficiency if inadequately treated, in which case being referred to as Hinman syndrome.

We present the case of a boy who developed full detrusor acontractility after undergoing a distal hypospadias repair, with no mechanical cause associated. As he was treated early by means of an appendicovesicostomy the development of hydronephrosis and of renal insufficiency that characterize Hinman Syndrome was possibly prevented.

Material and methods

Case report

A boy, born on 29/10/2008, of normal neuromotor development, with no cognitive deficit, and no difficulty to either urinate or evacuate. Healthy parents, living in harmony, he has a normal younger sister. In April 2011 he underwent a distal hypospadias repair. After the tube was removed he developed voiding difficulty, and had several events of urinary tract infection. A cystoscopy performed in August 2011 showed no point of stenosis or inflammatory reaction; normal urethra. Because he had events of urinary retention he first underwent a vesicostomy in September 2011 and, later, an appendicovesicostomy (Mitrofanoff principle) in January 2012. Actually, following the intervention for hypospadias repair he never again urinated normally, with urethral voiding only as he left the swimming-pool, when he then had a powerful, however very painful, urinary stream. The complete blood count, urinalysis, liver functions tests, renal function tests and blood electrolyte measurements were within normal limits. His renal ultrasonography was normal. A spine Magnetic Resonance performed in 2014 showed no alteration. In July 2016 he underwent ten sections of sacral neuromodulation with no change to bladder acontractility; he continued not voiding spontaneously and started having difficulty to evacuate. He had a MCUG in which the contrast was introduced through the appendicovesicostomy, but the voiding phase could not be obtained. He had a grade 1 vesicoureteral reflux on the left, with a

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Fig. 1. Cystography performed through the appendicovesicostomy shows smooth bladder wall and grade 1 reflux on the left.

bladder of normal capacity and smooth walls (Fig. 1). The ^{99m}TcDTPA-Renal Scan showed 24% function for the Left Kidney and 76% for the Right Kidney, both presenting non-obstructive curves (Fig. 2). A free uroflowmetry could not be performed at the urodynamic assessment since he failed to urinate spontaneously. A 15 mL urine residue was found at the start of the exam. He had 300 mL cystometric capacity, with good compliance. He presented desire to void at 130 mL, when he had detrusor hyperactivity. When reaching 300 mL despite marked voiding urgency he was unable to void; the bladder was emptied through the appendicovesicostomy (Fig. 3). Currently he undergoes urinary catheterization through the appendicovesicostomy, with no spontaneous voiding; he is being followed-up by a psychologist but so far, has been unable to void spontaneously.

Comment

Voiding is an extremely complex action that involves the sensitive nervous system, the voluntary motor somatic nervous system, the autonomic nervous system and the spinal cord and cortical coordination

centers, all acting simultaneously. Learning to void properly has to do with maturity of the muscles and nerves involved in the action, which may be influenced by the different psychological phases that the child is faced with along his/her development. Hinman and Bauman brought attention to the fact that a change to the normal psychic development may lead to so important a voiding disorder that it may cause hydronephrosis, resulting in renal insufficiency. There would be a detrusor hyperactivity that the patients would try to suppress by increasing the external sphincter.¹

The external sphincter contraction concept is currently accepted as being the cause of most voiding dysfunctions in children.² Fowler et al., in 1988 described a symptoms association, in which women, usually with Polycystic Ovaries, had acute urinary retention, whose substrate would be an abnormal electromyographic activity of the external sphincter.³ The disease, which became known as Fowler's syndrome, would be caused by a progesterone deficiency that usually occurs in women with polycystic ovaries. The progesterone stabilizes the cell membranes and its lack may allow for the ephaptic transmission of impulses among the muscular fibers, causing an abnormal electromyographic activity. Thus, different factors may influence the development of the voiding cycle, all of which, when the voiding dysfunction condition causes hydronephrosis and renal function disorder, are referred to as Hinman syndrome.

Voiding disorders are more and more recognized as being the cause of urinary infections and are associated with alterations such as vesicoureteral reflux and vaginal discharge in girls. The vesicoureteral reflux that develops in the antenatal period is currently recognized as a voiding dysfunction that, in most cases, disappears when alpha blockers are used. Therefore, one may say that bladder dysfunctions that occur due to detrusor hyperactivity during the bladder filling period, as well as those occurring during the bladder emptying period, may develop into a severe renal insufficiency condition and be labeled as Hinman syndrome. Our patient, following a surgical intervention that caused him a major trauma, stopped urinating through his penis, doing so only when the stimulus caused by the cold water of the swimming-pool triggered an involuntary and painful voiding, however, with a normal urine stream. Considering that he was treated early by means of a urinary diversion there was not time enough for the renal function to be severely affected.

Increasingly recognized as the cause of urinary disorders, the dysfunctions ought to be recognized as early as possible, and treated whether with drugs, physiotherapy or, occasionally, with psychological follow-up, as many times, when not recognized and therefore, not treated, they may hinder the proper development of the voiding capacity, case in which they are referred to as Hinman syndrome.

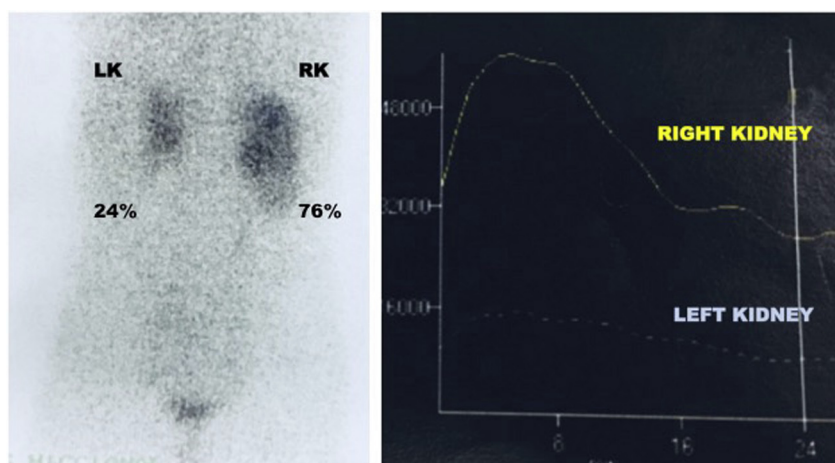


Fig. 2. Dynamic ^{99m}Tc DTPA scintigraphy showing curves of the non-obstructive type for both kidneys, with distribution of the radiotracer at 24% for the left kidney and 76% for the right kidney.

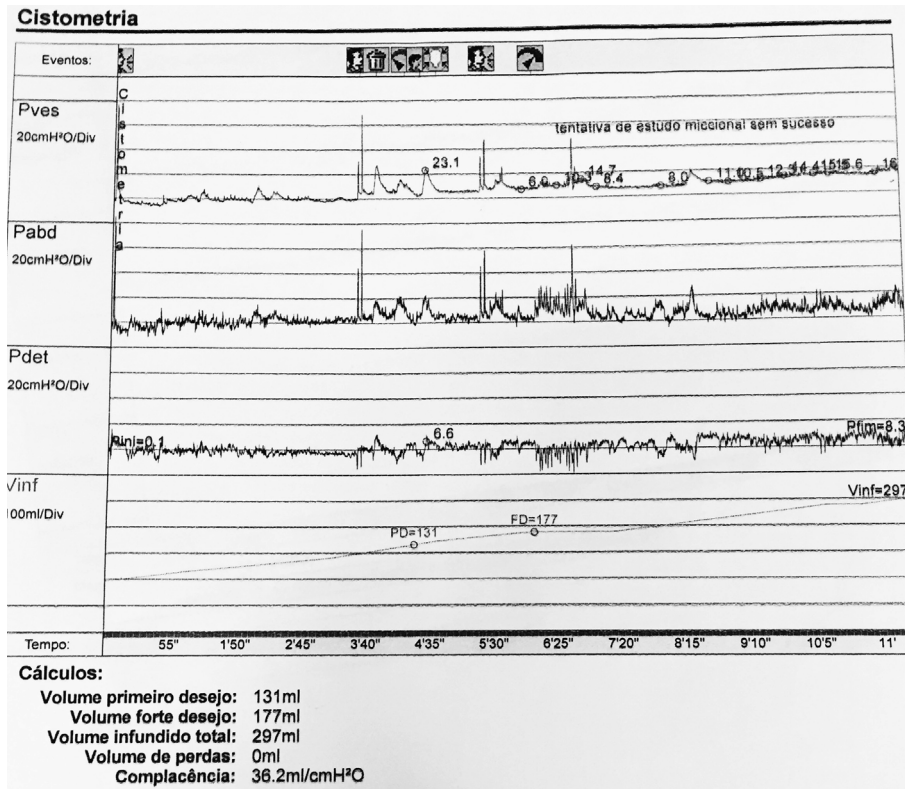


Fig. 3. Urodynamic assessment showing detrusor hyperactivity and acontractility.

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