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Hutch diverticulum: from embryology to clinical practice

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Abstract: Hutch Diverticulum (HD) is defined as the protrusion of the mucosal and submucosal layer through the muscle bundles of the underlying detrusor muscle. HD is located at the vesicoureteral junction with a backward direction from the homolateral ureteral orifice. As far as its etiology is concerned, HD is caused either by a congenital muscle wall defect at the level where the Waldeyer's fascia occupies the clefts between the vesical part of the homolateral ureter and the detrusor, or is associated with abortive ureteral duplication or defective incorporation of mesonephric duct into the bladder at the site of ureteral hiatus or finally is associated with the development of transient urethral obstruction. HD is usually unilateral and more common in male patients. It may be associated with the Ehlers-Danlos, Williams-Elfin and Menkes syndromes. HD usually occurs in childhood and rarely during adulthood. It is found in 0.2–13% of all children presenting with urinary tract infection. Through this short review article, we attempt to present in detail the most recent bibliographic data concerning this entity, focusing on pathophysiology, diagnostic approach, and treatment strategy.

Keywords: congenital, diverticulum, paraureteric (Hutch), Waldeyer's fascia, ureterovesical junction, vesicoureteral reflux, child.

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Introduction

Hutch Diverticula (HD) are very rare, affecting mainly male patients, with a male to female ratio equal to 9:1 [1, 2]. They are defined as the herniation of the mucosa and submucosa of the urinary bladder through fine muscle fibers of the detrusor muscle. HD usually present during childhood and rarely in adulthood (66% vs. 34%) [3, 4]. They are divided into true, which are more frequent, and false. True are characterized

by the spherical herniation of the bladder mucosa to the adjusted muscular wall, whereas the false are characterized by herniation of the bladder mucosa between the detrusor muscular fibers.

In a previous retrospective analysis by Garat *et al.* concerning 10 cases of surgically treated HD it was shown that, after histopathological examination, diverticulum's epithelium was covered by fine smooth muscle fibers. False HD do not feature muscle fibers, whereas true HD consist of mucosa internally and fibers of the detrusor muscle externally [2].

In contrary to this terminology, we refer to the secondary diverticuli developing as a result of increased endocystic pressure during both the resting state and micturition. This is a result of either a subcystic obstruction or the presence of neurogenic bladder. Those HD located at the ureterovesical junction are defined under the term "congentital paraureteric diverticuli" [5].

HD are usually unilateral. In 8% of all cases, an association with Ehlers-Danlos syndrome is identified. They can also be related to various syndromes such as Prune-Belly, Williams and Menkes [6–10]. They are usually found posterior and lateral to the adjacent ureteral orifice [5].

Embryology

HD can form as a result of a congenital insufficiency of the muscular wall where the Waldeyer's peritoneum fill the gap between the wall of the ureter and the detrusor muscle. On this basis, increased endocystic pressure has a catalytic effect. Subsequently, a limited part of the bladder mucosa protrudes through the gap. Progressively, and in the presence of increased endocystic pressure, the gap widens leading to a protrusion of a larger part of the mucosa. Finally, the ureteral orifice is incorporated into the HD wall, resulting in ureterovesical junction insufficiency and finally in vesico-ureteral reflux (VUR) [11].

HD can be attributed to abortive ureteral duplication. It can also be related to defective incorporation of the mesonephric duct into the wall of the urinary bladder at the level of the ureteral hiatus [12]. HD can be related to the development of intermittent subcystic obstruction, due to the angular distortion of the developing urethra or its obstruction by a comper gland in the presence of a membrane within the duct of the posterior urethra [13].

Pathophysiology

Retention of urine within the HD could result from a narrow neck or from the lack of support to the corpus by the muscular fibers of the detrusor muscle. Urine retention is therefore responsible for recurrent urinary tract infections or genesis

of lithiasis, whereas malignancy of the cystic wall can also develop due to chronic irritation [1, 14, 15].

The retention of urine in the HD leading to its progressive dilatation, along with the subsequent involvement of the adjacent ureter within the wall of the diverticulum account for the insufficiency of the ureterovesical junction. This may prolong or worsen the VUR. An obstruction at the level of the ureterovesical junction may also develop [1].

When the HD reaches great size, it can extend extravesically, towards the bladder neck, acting like a subcystic obstruction. If the HD is characterized by a wide neck, paradoxical movement of the urine may occur (urine directing towards the diverticulum and not the urethra) during the contraction of the detrusor muscle. This dysfunction exhausts the developing pressure for urine extraction, finally resulting in increased work and double urination.

Clinical presentation

According to the pathophysiology, main clinical manifestations of HD include recurrent urinary tract infections, urolithiasis and urinary defects, namely vesicoureteral reflux or obstruction at the level of the uterovesical junction [16, 17].

Garat *et al.* summarized main symptoms in their retrospective analysis; febrile urinary tract infections (5/11 cases), recurrent urinary tract infections (3/11 cases), enuresis (1/11 cases), macroscopic hematuria (1/11 cases) and abdominal pain (1/11 cases) [2].

During diagnostic approach of children with urinary tract infections, HD is recognized as the underlying cause in 0.2–13% of all cases [18]. Description of symptoms such as double urination or limited width of urinary stream can lead to the diagnosis of concomitant VUR [11].

Concerning other, less frequent clinical manifestations, Singh *et al.* described a case of acute urinary retention in a newborn with HD [11]. Oye *et al.* also referred to a boy with acute urinary retention due to obstruction of the urinary bladder's neck from the extravesical extension of a large HD [19].

Relevant systematic search in MEDLINE between 1970 and 2012 resulted in the identification of 53 cases of acute urine retention due to HD. In summary, 33/53 cases were described in neonates, 16/53 referred to children, whereas only 2 cases involved adults [11]. This specific complication is the more frequent during neonatal period, because the prostatic part of the posterior urethra is surrounded by loose connective tissue. Thus, it can be displaced in front of the posteriocystic protruding HD [20].

Abdullah *et al.* described an interesting case of a female adult patient reporting symptoms finally attributed to the exterior obstruction of the rectum by a large HD [21].

Rarely, a HD can present with established chronic kidney disease, as a result of multiple episodes of acute pyelonephritis, intrarenal reflux or obstructive uropathy [7, 8, 22]. Finally, HD might also be asymptomatic during childhood, identified later, during adulthood [15].

Diagnostic approach

The main diagnostic modalities include ultrasonography of the genitourinary system, antegrade cystourethrogram and dynamic tests including manometry and urometry. Ultrasonography is a key imaging study in order to determine the potential consequences of VUR and, rarely, of obstructive uropathy, and evaluate any renal or ureteral damage. Furthermore, it is utilized to evaluate the vesicourethral functional unit; the anatomy and activity of the detrusor muscle, the bladder neck, the ureteral diameter at the ureterovesical junction and the volume of urine produced in storage phase and after voiding. The evaluation of the volume of residual urine in the bladder after urination is of great importance. Lastly, it is considered as necessary in order to evaluate the anatomic behavior of a large HD during the two phases, storage and voiding.

The antegrade cystourethrogram, which is the major diagnostic modality, visualizes both the storage and the voiding phase of the bladder [11, 23]. This is appropriate for the proper and thorough evaluation of the neck of the bladder and the urethra. The dynamic testing of the vesicourethral functional unit with the use of manometry and urometry is only indicated in children older than 4 years of age, when a neurogenic bladder or a functional bladder defect is highly suspected.

Before surgery, endoscopy of the lower genitourinary tract could be helpful in determination of the location of the diverticulum and its anatomical position in relationship with the adjacent ureter, the width of its neck, evaluation of the urethra, bladder and the morphology of the ureteral orifices [11].

Operative indications

A gigantic diverticulum, sizing more than 1/3 of the urinary bladder transverse diameter, producing major complications such as a recurrent urinary tract infection, lithiasis, subcystic obstruction and rupture, are considered as indications for surgical management [15, 16, 19, 24–26]. During childhood, main indications are the coexistence of vesicoureteral reflux and the obstruction of the uterovesical junction [15, 24].

Gingh *et al.* and Garat *et al.* believe that in the coexistence of HD of small diameter (<2 cm) and VUR, the possibility of its regression is similar with the VUR cases without the presence of a diverticulum; thus, surgical treatment

is not suggested. Therefore, they specify the indication of surgical intervention in cases when VUR exists with an underlying HD, greater than 2 cm in diameter [11, 27].

Principles of surgical management

The classic surgical approach of removal of a HD is via cystectomy. Nowadays, the excision of a HD can be achieved transurethrally, laparoscopically and robotically assisted [18, 28]. The transurethral method is considered as the treatment of choice for a small HD, performing either a resection at the level of the neck or an inversion towards the bladder mucosa [11].

In the context of the open method, the catheterization of the ipsilateral ureter is the first step and then, the mural part is prepared. Next, the diverticulum's mucosa is removed, and the underlying muscular wall is reinforced. If the ureter is part of the wall of diverticulum, resection of the diverticulum and reimplantation of the ureter are performed. In cases when HD has small dimensions, all the surgical procedures are performed intravesically, whereas, if the HD is large, a combined intravesical and extravesical approach is considered as necessary [2, 11].

In cases of a large HD with urgent symptomatology during the neonatal period, treatment requires 2 phases: a cutaneous vesicostomy precedes, and in average, after one year the main procedure is performed [7].

Levard *et al.* refer to the defect of the muscle sublayer of the cystic wall as a causative factor for the most common postoperative complication of HD in children with Ehlers-Danlos syndrome [16].

In the context of the procedure two major issues arise:

— during the removal of a HD, the intravesical part of the ureter must be preserved
— during the reinforcement of the cystic wall after the removal of a HD, attention must be paid in order to avoid injury of the adjacent vas deferens.

Conflict of interest

None declared.

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