International Journal of Urology (2003) **10,** 459–462

Case Report

Papillary adenoma of the prostatic urethra: Report of two cases

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Abstract Papillary adenoma of the prostatic urethra is a rare cause of hematuria, hemospermia and lower urinary tract obstruction, especially in young adults. We report two cases of papillary adenoma and their endoscopic views. One of the patients had a lesion located on the verumontanum and presented with hematuria, hemospermia and symptoms of lower urinary tract obstruction. The other patient had papillary lesions located in the paramontanal gutter and presented with persistent painless hematuria. The origin of both lesions was proven to be prostatic epithelial differentiation using immunohistochemical identification of prostate-specific antigen. Papillary adenoma is believed to be benign, therefore, both lesions were carefully excised and fulgurated transurethrally.

Key words hematuria, hemospermia, lower urinary tract obstruction, papillary adenoma, prostate, urethra.

Introduction

Polypoid lesions of the prostatic urethra are occasionally observed in patients with hematuria and/or hemospermia. Papillary adenoma is a rare lesion of prostatic origin. The condition has many synonyms, including 'ectopic prostatic tissue in the urethra', 'benign polyp of prostatic-type epithelium', 'prostatic caruncle', 'adenomatous polyp of the prostatic urethra', 'prostatic urethral polyp', 'benign prostatic epithelial polyp' and 'papillary adenoma of the prostatic urethra'.[1–8] Although the lesion is well-described, due to the unfamiliarity with its endoscopic appearance it could easily be ignored or missed in young adults presenting with hematuria, hemospermia and symptoms of lower urinary tract obstruction. We present in this paper two cases of papillary adenoma and their endoscopic views.

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Received 31 May 2002; accepted 20 January 2003.

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Case 1

In October 2000, a 35-year-old man presented to our institution with gross hematuria, hemospermia and a 6-month history of lower urinary tract obstruction symptoms. The patient was treated with three courses of antimicrobial therapy but his symptoms persisted. Laboratory test results were all within normal limits. The patient's urine sediment showed an abundance of erythrocytes. On excretory urogram, the upper urinary tracts were found to be normal. Transrectal and pelvic ultrasound examinations were also normal. On urethroscopy, the verumontanum appeared to be hypertrophied, edematous and blood-stained, and it obstructed the prostatic urethra (Fig. 1). Transurethral resection of the hyperplastic verumontanum was performed under lumbar anesthesia.

Case 2

In December 2000, a 40-year-old male presented to our institution with an 18-month history of gross, painless and persistent hematuria. Physical examination results

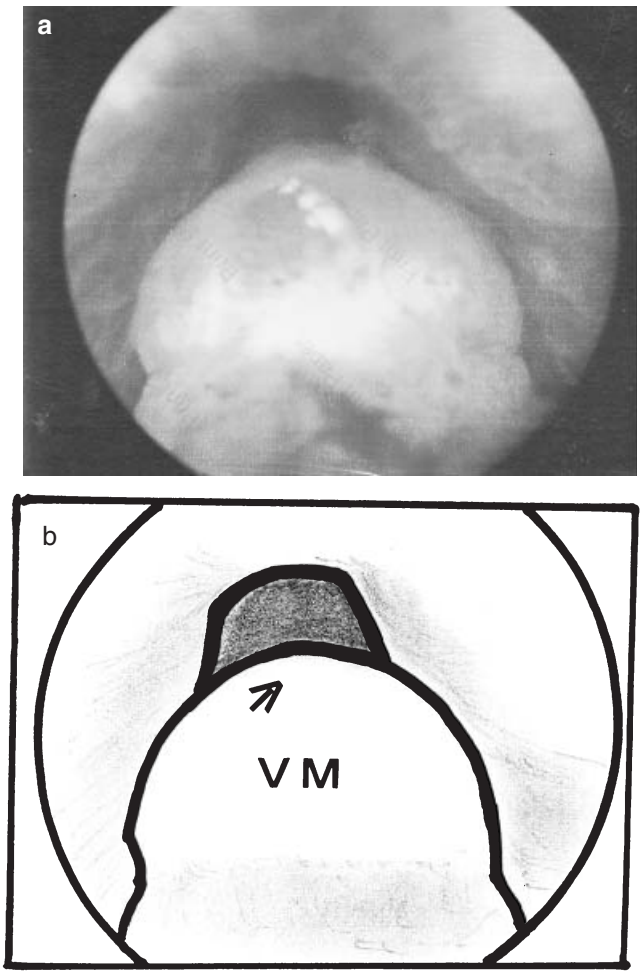


Fig. 1 Papillary adenoma in Case 1. (a) Endoscopic appearance. (b) Schematic presentation showing the papillary adenoma (arrow). VM, hypertrophic verumontanum.

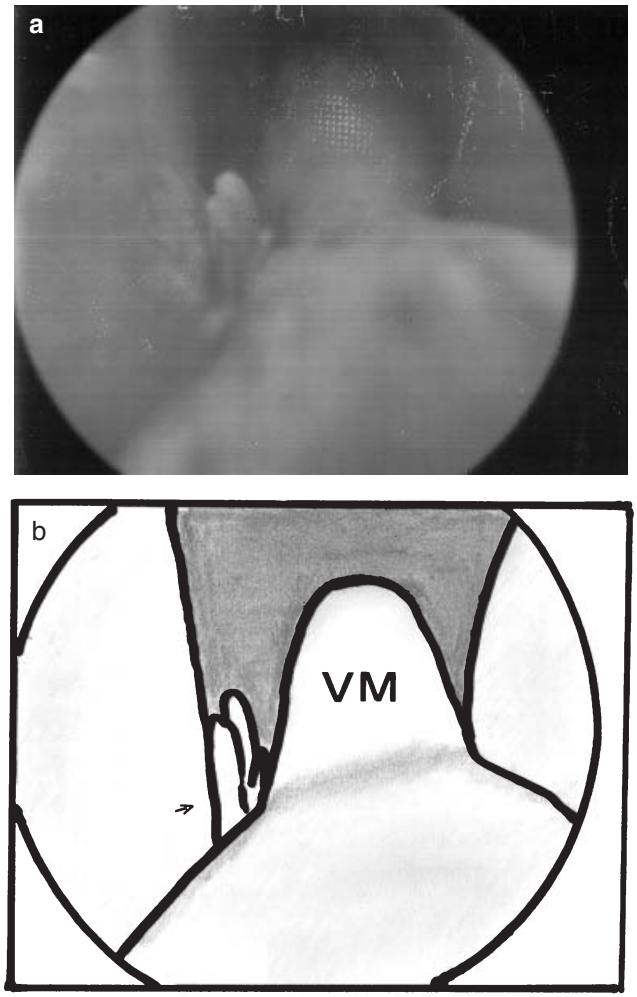


Fig. 2 Papillary adenoma in Case 2. (a) Endoscopic appearance. (b) Schematic presentation showing finger-like polyps (arrow) rising from the paramontanal gutter. VM, verumontanum.

were normal and routine laboratory tests were within normal limits. Urine analysis showed 20–25 erythrocytes per high power field. On excretory urogram, the upper tracts were found to be normal. Cystourethroscopy revealed finger-like polyps rising from the paramontanal gutter (Fig. 2). Transurethral resection and fulguration of the polyp bases were performed under lumbar anesthesia.

Microscopically resected polyps consisted of a surface epithelium lined by two layers of cells forming papillary gland-like structures. The luminal layer comprised tall columnar cells resembling normal prostatic acinar cells of eosinophilic cytoplasm and basally located nuclei. An underlying cuboidal cell layer was present. There was no evidence of cellular pleomorphism or dysplasia and mitosis (Fig. 3a). Immunohistochemical identification of prostatic-specific antigen using the peroxidase–antiperoxidase technique (Fig. 3b) proved the origin of both lesions to be prostatic epithelial differentiation. Thus, the diagnosis was papillary adenoma.

Both patients were followed-up by repeat urinalyzes and endoscopic evaluation at 6-month intervals after the resection. The first patient was followed-up for 16 months and the other for 18 months. Endoscopically, both patients have remained free from relapse during these periods. Hematuria and hemospermia had disappeared in both patients and symptoms of lower urinary tract obstruction were resolved in patient 1.

Discussion

Polypoid lesions of the prostate were first described and classified by Randall in 1913.[9] Although they have

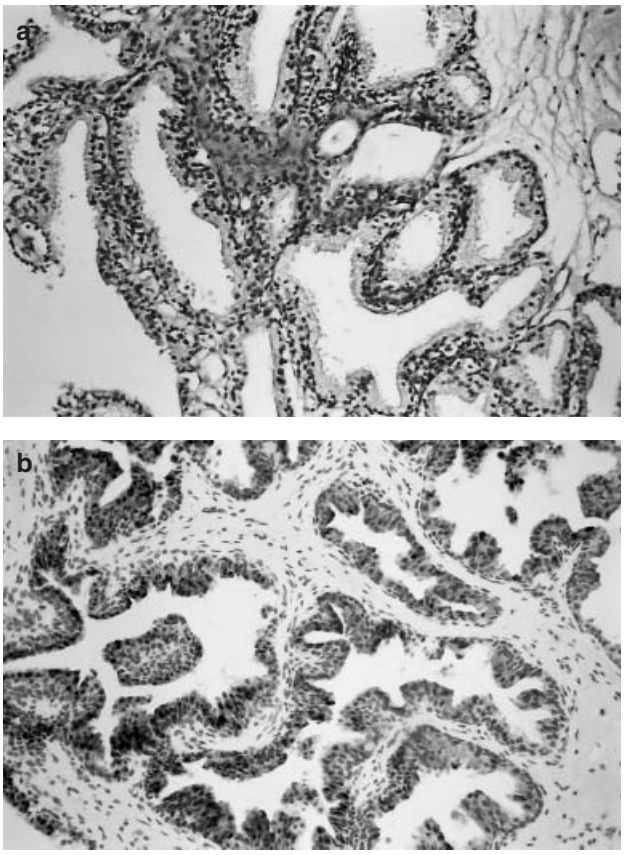


Fig. 3 Histological examination of papillary adenoma showing papillary formation of the surface epithelium and glandular lining. The acinar cells were arranged in two layers: a luminal layer of tall columnar epithelium; and (a) the underlying cuboidal cell layer. (b) Diffuse positive prostate-specific antigen staining (H&E × 200).

similar endoscopic features, their origins are quite different. Polyps in infants and young children are of congenital origin while polypoid urethritis is the counterpart of inflammatory cystitis.[8] Adenomatous polyps of prostatic type epithelium represent a separate clinicopathological entity. In 1962, Nesbit was the first to realize the histological similarity of the lesion with prostate acini by gland-like structures covered with a papillary columnar epithelium.[10] Inflammation may not present characteristically but prominent submucosal capillaries are a common feature. Through a series of 12 cases, he presented evidence that the epithelium stained positively with azo dye techniques, suggesting prostatic acid phosphatase content. In 1971, Butterick *et al*. examined the same lesions under electron microscopy and reported that the lesion has similar ultrastructural features to the prostate gland.[4] Walker *et al.*,[2] and later, Eglen and Pontius[6] demonstrated the presence of prostatic acid phosphatase as well as prostate-specific antigen, further confirming the prostatic origin of the lesion. Most authors believed that the lesion represented ectopic or aberrant prostatic tissue.[4,6,10] However, other mechanisms have been suggested, such as a benign neoplasm of the prostate epithelium projecting into the lumen of the urethra or a defect in the embryologic invagination process of submucous glands of the inner zone of the prostate.[6,10,11] Endoscopically, the lesions are most commonly located in the paramontanal gutter in 76% of the cases, followed by the verumontanum in 20%.[11] The lesions are usually single but can be multiple or diffuse. They are usually papillary or polypoid but can be sessile or villous.[6] Occasionally, no localized lesion is seen but there is a diffuse thickening of the anterior mucosa covering the verumontanum and adjacent areas. These cases are the most difficult to recognize and are often missed or ignored as in our case. In other cases, the mucosal surface is granular and velvety.[12] Examples of the usual endoscopic appearance were rarely found in the literature.[8]

Adenomatous polyps of prostatic type epithelium have numerous synonyms as stated above.[1–7] Although the main pathological process is the presence of ectopic prostate tissue in the urethra, some authors prefer to use the term 'benign prostatic epithelial polyp' because of its endoscopic appearance,[6,8] while others have given it the name 'papillary adenoma'.[11,12] Malignant transformation of an adenomatous polyp is very rare, and to our knowledge, only one case has been reported.[13] Walker *et al.* reported a case of endometrioid carcinoma of the prostatic urethra arising from a villous polyp,[13] and in 1983, the same authors reported six cases of prostatic type polyps, one of which contained foci of stratified, mitotically active cells.[2] This suggests that these polyps may represent the benign counterpart of endometrioid carcinoma of the prostatic urethra and have the potential for malignant transformation. We agree with Baroudy and O'Connel,[11] and Zeid *et al*.[12] that the lesion has a neoplastic potential and does not always manifest itself as a polypoid structure, hence the term 'papillary adenoma' would be more appropriate.

Papillary adenomas of the prostate usually occur in young adults. Although the actual age range extends from 13 to 70 years[12] they have never been reported to occur before puberty. A series of patients reported from Japan by Hara and Horie[5] and one from Saudi Arabia by Baroudy and O'Connel[11] demonstrate the lack of any specific geographic preponderance. Initial gross or microscopic hematuria and/or hemospermia are the usual clinical symptoms, especially in young adults. Baroudy and O'Connel reported that papillary adenoma of the prostatic urethra was the most common cause of hemospermia and hematuria in young men in Saudi

Arabia.[11] Papillary adenomas are extremely rarely reported to cause urinary obstruction symptoms, therefore, our first case could be an important contribution to the literature.[8,12] Goldstein *et al*.[14] also reported that this lesion might be more frequent than is usually expected. We share the view of Goldstein *et al*. that this lesion is ignored or missed in especially young cases who present with hematuria and/or hemospermia and is, thus, generally accepted as idiopathic. Therefore, familiarity with its usual endoscopic appearance could help urologists easily recognize and treat this lesion.

Transurethral resection and fulguration have provided an adequate therapeutic approach in most cases, however, considerable caution must be taken because of the anatomic proximity of the external sphincter to the verumontanum.[15] Recurrent development of these lesions after local resection is unusual[4] but several cases have been reported.[1,15] Due to the rarity of the lesion, debate continues on the prognosis, however, regular follow-up is recommended.

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